THE
BRITISH JOURNAL OF SURGERY

VOL. XXXVI JULY, 1948 No. 141

HYPERPLASTIC CALLUS FORMATION, WITH OR WITHOUT EVIDENCE OF A FRACTURE, IN OSTEOGENESIS IMPERFECTA

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IN 1943 Brailsford, in an article on osteogenesis imperfecta, referred to a complication which he described as "a condition suggesting scurvy, subperiosteal hæmorrhages being more frequent than fractures", and he added brief notes of 4 cases.

In 1937 Dr. W. Mackenzie sent me the notes and radiographs of a case, apparently one of osteogenesis imperfecta—a lad of 14, with a curious enlargement of the lower half or more of the shaft of one femur, and bony excrescences on the forearm bones. Since that date other friends have provided me with details of 7 more or less similar cases, 3 of which I have had the opportunity of examining. Most, if not all, of these cases are undoubtedly similar to those recorded by Brailsford; in fact, two of Brailsford's cases are included in my series, the surgeons in charge having also sent me radiographs and detailed notes of their respective cases (*Cases* 1 and 3).

It is the purpose of this paper to call further attention to a curious and obscure condition and to record the chief features of the individual cases included in this series. The aetiology suggested by Brailsford will be questioned, since the evidence that scurvy is partly responsible for the curious features of these cases is by no means convincing.

The thickening of the bones here considered is quite distinct from the enlargement of the shafts of certain of the long bones met with in the first few months of postnatal life in cases of osteogenesis imperfecta of antenatal origin (Fairbank, 1927, 1930).

The formation of callus after fracture in ordinary cases of osteogenesis imperfecta is usually, but not always, plentiful, and sometimes rather excessive, but the excess is absorbed in the normal way as consolidation occurs. Examination of our records of some 40 other cases of osteogenesis imperfecta has revealed none in which callus formation was in the least comparable with that seen in the series under discussion.

Of the present series only 6 appear to be cases of osteogenesis imperfecta, all apparently of the postnatal type, and in 2 of these the evidence is not very strong. All 6, however, showed general osteoporosis of the skeleton, with a history of a varying number of fractures up to "nearly every bone in the body". In the remaining two that are not regarded as osteogenesis imperfecta, there is no history of multiple fractures, but one of these had definite signs of generalized osteoporosis, and both had had a single fracture, of the ulna in one, and of the ankle in the other. In no case did the sclerotics display the deep blue colour so striking a feature in some cases of osteogenesis imperfecta. The features displayed in this series are:—

a. Formation of intensely calcified local callus, not invariably preceded by a recognizable recent fracture.

b. Excessive formation of ossified callus, which enveloped the shaft to a varying but quite unusual extent, and in some cases displayed a strong tendency to result in permanent enlargement of the affected bone. Evidence of antecedent fractures of the affected bones was again not invariably apparent in the radiographs.

c. Formation of bony excrescences on the shafts of long bones, particularly on the interosseous borders and without the faintest sign of antecedent fractures.

Excessive formation of *early callus of unusual density* was noted in 3 cases, all apparently suffering from osteogenesis imperfecta. (*Cases* 2, 3, and 4.) In two of these the callus formation followed a fracture, while in the third swelling of the part and the appearance of the dense shadow undoubtedly occurred on at least some occasions without the faintest sign of fracture. The dense and sometimes completely structureless shadow produced by what appears to be excessive calcification of the callus in these cases bulges out on one or both sides of the bone almost like the wings of a butterfly, a rather grotesque appearance quite unlike the faint fusiform shadow cast by normal callus in the early stages of its ossification. In each case a femur was the bone affected in this way, but in one a humerus, without a fracture, was involved in addition. The femur in each case later developed extensive masses of callus, with permanent enlargement of the shaft of the bone. The dense callus on the humerus was later absorbed.

Excessive formation of periosteal bone, usually starting from a fracture and therefore legitimately

regarded as callus, was a definite feature of 7 of the cases, and even in the remaining case (*Case* 7) there were some signs of this in one femur. It was with some hesitation that this last case was included in the series. The femur is the chief bone affected, this bone being involved in all 7 cases, while in 6 of these both femora were involved. The new bone extends far beyond the region of the fracture, when one is visible, and involves a length of the shaft that could not possibly have been affected by trauma at the time of the injury. At first the original cortex of the bone is easily seen within the sheath of new bone, but later this may be absorbed. The affected portion of the shaft, half its length or more, may remain permanently enlarged to twice its normal size or even more, the enlarged portion consisting of open-mesh or coarsely cancellous bone. In *Case* 2, undoubtedly one of osteogenesis imperfecta with hyperplastic callus of the left femur, later films of the right femur show a curious abrupt fusiform enlargement in the upper half of the shaft rather suggestive of a cyst, with an abrupt irregular-shaped mass of callus projecting inwards from its surface.

Both humeri were affected in 2 cases: in one of these it was quite obvious that no fracture had occurred. The other long bones are much less often affected: in 2 cases several bones were involved to some extent. It is noteworthy that though excessive callus formation has followed a fracture of a femur, fractures of other bones, even in the same leg, e.g., *Case* 4, have united in a perfectly normal manner with no excess of callus. In *Case* 1, with marked enlargement of one femur following a "contusion", the opposite femur had been broken, in addition to other bones, but this femur had united with no more than a normal amount of callus. Ten years later, however, this second femur suddenly flared, an enormous mass of callus developing rapidly around the upper third of the shaft: there was no history of recent injury and no sign of a fracture. In some cases, even in spite of splintage, the swelling has progressed and involved the soft tissues so extensively, with enlargement of the superficial veins and occasionally redness of the skin, that a suspicion of malignant growth has arisen. This has resulted in the performance of a biopsy in 4 cases—in all of which the femur was the bone affected. The fact that one of these cases was under the care of my friend Professor Harry Platt, whose experience of bone tumours is quite exceptional, is significant. In no case in this series has the suspicion of malignancy been confirmed. Brailsford reports the appearance of "malignant metaplasia" involving a tibia in one of his cases, but he does not state whether or not this was confirmed by biopsy.* In 2 cases osteotomy was performed for the correction of deformity and in one of these some bone was removed for microscopical examination.

Bony excrescences were present in at least 6 of the cases. The radius and ulna were the bones most frequently affected, the shafts of these bones showing projections, usually on the interosseous border, in 5 of the cases. In shape they varied from rounded to spicular. In 1 case the excrescences had fused and there was synostosis of the two bones in one forearm. Occasionally the additional bone was so smooth and flat it suggested the simple cortical thickening seen after union of a fracture; but no sign of a fracture, recent or old, was visible, and the extra bone was seen on one side only of the shaft. The other bones showing bony excrescences in one or more cases were the pelvis, the femur, the humerus, the tibia, and the fibula, particularly the fibula. Only in 1 case did the metacarpals and metatarsals show some ridges. In no case was there clear evidence of an antecedent fracture in the neighbourhood of these excrescences, though the possibility of a fracture at some time in the past could not be entirely excluded. It seems probable that some at least of these thickenings and projections had resulted from trauma that failed to produce a fracture. Occasionally it was not easy to decide whether the excess of bone should be classified as hyperplastic callus or simply as an excrescence.

Other points worthy of mention in the series are the following: The ages at which the cases first showed signs of a tendency to excessive callus formation varied from one to fifteen years. *Case* 1 had reached the age of 24 years before the second femur was affected: gross enlargement of the femur first affected—ten years previously—was still present. In another case the hyperostosis of certain bones was still present at 25. Five were males, three were females. In six growth was stunted: the remaining two, though undoubtedly suffering from osteogenesis imperfecta, were of normal height. Only in 1 case (*Case* 6) was the family history, reported by Dr. Hilton (1934), of special interest, the mother and two maternal aunts being stunted in growth and having changes in the bones said to be similar to those seen in the young woman reported, while a male cousin was said to have deformed arms, possibly similar to those of the patient, who had bilateral dislocation of the radial heads.

Curvature of the bones and deformities of the limbs, the result of bending or fractures, were present in all cases, the legs and forearms being particularly affected. Contracture of the hips and knees was present in 1 case, of the hips alone in 1, and of both elbows in 3 cases. In each of the latter three there was bilateral dislocation of the radial heads, a curious and rather surprising complication: in one of these cases the wrist movements were also limited. The dislocation is apparently secondary to bending of the forearm bones and shortening of the ulnæ. The pelvis was indented in 1 case, and the acetabula unduly deep, while in another definite protusio acetabuli was present bilaterally, with a crack in the floor of the acetabulum on one side. In this case the ischia were curiously enlarged. The skull showed nothing that osteogenesis imperfecta could not account for. In no case were the epiphyses involved in any of the changes described above, with the exception of osteoporosis, but in 2 cases slipping of the femoral heads had occurred, in one hip only in one case, and in both hips in the other. In 5 cases the spine showed osteoporosis, with bi-concave vertebral bodies, as commonly seen in osteogenesis imperfecta. Though scoliosis was present in 2 cases, and kypholordosis in another, the spine displayed nothing of special interest.

* Professor Baker heard from Dr. Brailsford recently that this case died with metastases.

Blood examination in every case revealed nothing abnormal or helpful. The high phosphatase reported in 3 cases could easily be explained by recent fractures. A definite report on the Wassermann reaction was obtained in only 3 cases, being negative in each. Petechial hæmorrhages in the skin over an unprovoked swelling of an elbow were found in 1 case (*Case* 3): this case also had one mild attack of nose-bleeding.

As to treatment, apart from that called for by fractures, all these cases had spent considerable periods, often extending to years, in special children's or other hospitals, where they must have had more than adequate dietetic treatment. Any possible deficiency of vitamin C must have been met, and yet some of the cases developed the curious features described above while actually in hospital. It is known that in 2 cases intensive treatment to correct a possible deficiency of vitamin C was carried out over prolonged periods without appreciable effect on the condition of the bones.

CASE REPORTS

The following are the more important details of the individual cases :—

Case 1.—H. E., male, aged 14 (1937). History of multiple fractures involving both thighs and both arms. Right thigh contused when 7, and the enlargement noticed a year later: this is still present. At one time amputation advised. No fractures for past six years. No family history of similar condition, but two brothers sustained fractures in childhood. Patient below average in height. Lower half of right femur generally thickened.

FIG. 1.—*Case* 1. Anteroposterior and lateral views of right femur showing the enlargement of the lower half of the shaft to be due to the addition of bone outside the cortex. Film taken (June, 1937) six years after the enlargement was first discovered.

This limb longer than left, the femur being chiefly responsible. Curvature of radius and ulna in both forearms. Marked occipital and some supra-auricular bulging of the skull. Sclerotics not unduly blue. Blood examination revealed nothing important, though plasma-phosphorus was low and phosphatase high.

X-ray Examination.—Osteoporosis was not a striking feature of skeleton as a whole, but was present in the extremities of long bones, and markedly in spine, the vertebral bodies being typically shallow, spread, and biconcave. Lower half of shaft of right femur showed fusiform enlargement to about three times normal size, the additional bone being honeycombed. In lateral view cortex of shaft was clearly seen with new bone outside it. No sign of an old fracture. Surface of additional bone was for the most part smooth and gave impression of permanency (*Fig.* 1.) There was an old healed fracture of left femur, left clavicle, and possibly one rib, all without excess of callus. Some irregular bony excrescences along interosseous borders of upper half of both radii and a trifle of one ulna. Clear cortical lesion (? chondroma) seen in proximal phalanx of left fourth finger. Bones of vertex of skull unduly thin.

FIG. 2.—*Case* 1. Left femur (Jan., 1948) showing mass of callus five weeks after onset of pain and swelling. No sign of fracture.

In December, 1947, he complained of sudden pain and swelling of left thigh. No history of recent injury. Twelve days after the onset the circumference of the thigh was 4 in. greater than right. Radiographs showed large mass of callus—not particularly dense—around upper third of the femur. No sign of fracture. Five weeks after the onset the mass of callus was enormous, the total diameter of the shadow of the callus amounting to 5 in. (*Fig.* 2). Films of right femur showed that little if any change had taken place during the past ten years.

Biopsy specimen sent to Professor Baker, who reports " a mass of pale semicartilaginous tissue which on section showed chondroid tissue and cartilage similar to the appearances seen in *Cases* 4 and 8 ". (A further report with detailed histological findings will be published later.)

(Under Dr. Mackenzie.)

Comment.—It seems justifiable to regard this case as one of osteogenesis imperfecta of a mild type. It is noteworthy that while one femur was affected by hyperplastic callus, with every appearance of this being permanent, fracture of the left femur had resulted in no excess of callus. Yet ten years later a large mass of hyperplastic callus formed suddenly about the left femur, with no provocative fracture.

Case 2.—P. W., male, aged 4 years (1938). History of multiple fractures affecting at least eight bones since age of 6 months, and of deformities resulting therefrom. No other member of family affected. Sclerotics bluish, but not typical.

X-ray Examination.—Generalized osteoporosis, and clear signs of old fractures in one femur, both tibiæ and fibulæ, and left humerus. Many bony excrescences along

FIG. 3.—*Case* 2. Left elbow (Sept., 1938) showing excess of callus following fracture of the lower third of humerus, and abnormalities of the bones forming the joint.

interosseous borders of right radius and ulna, which were curved: the radial head was displaced outwards. Similar but less marked hyperostosis in the left forearm and on one fibula. Excessive callus surrounding fracture of lower third of left humerus and smooth palisade-like bony outgrowth from outer supracondylar ridge of right humerus (*Figs.* 3, 4). In 1940 excess of callus had disappeared from left humerus, leaving slight fusiform enlargement. In January, 1942, without fracture, right ulnar shaft showed excessive bone formation on outer side, the additional bone being of uniform low density, and with no sign of trabecular structure (*Fig.* 5). At this date the major long bones had slender osteoporotic shafts typical of osteogenesis imperfecta. Left femur showed recent fracture in upper half with excess of dense callus around it and less dense callus extending down the shaft to an unusual extent (*Fig.* 7). In February, 1942, osteotomy for correction of deformity of left femur gave opportunity for biopsy of callus. Histological report—" Considerable osteoblastic and some osteoclastic activity. There is also formation of coarse fibrous bone, apparently by metaplasia of cartilage on surface. No osteoid seams seen ". Four months later callus still excessive, but much less dense and more diffuse (*Fig.* 8). Blood examination revealed nothing of importance. In 1943 the right femur also fractured: the early callus, though not excessive, was unusually dense. Large doses of vitamin C given without

FIG. 4.—*Case* 2. Right arm (Sept., 1938) showing abnormal formation of bone, without fracture, along the lateral supracondylar ridge, displacement of head of radius, and deformity of the radius and ulna with many bony excrescences along their interosseous borders.

effect. In 1945 grotesque irregularity in shape of left radius and ulna with marked dislocation of head of radius. Some bony outgrowth from lower third of humerus. Upper third of left femur still enlarged, generally honeycombed, and poorly calcified. At site of old fracture of right femur was a fusiform enlargement consisting of honeycombed bone with a mass of bone projecting abruptly inwards for over an inch. Both femoral heads had slipped, the necks being abnormally small. In 1946 marked limitation of movement of elbows and forearms. Could just walk with assistance: never walked at all till 10 years old, and then only with appliances and assistance.

Scoliosis present. Sessile poorly calcified projection from cortex on outer side of shaft of right humerus. Right ulna showed further enlargement on outer side, the excess of bone being almost entirely structureless and of uniform low density: radial shaft showed signs of absorption from pressure by the enlarged ulna. In 1947 some

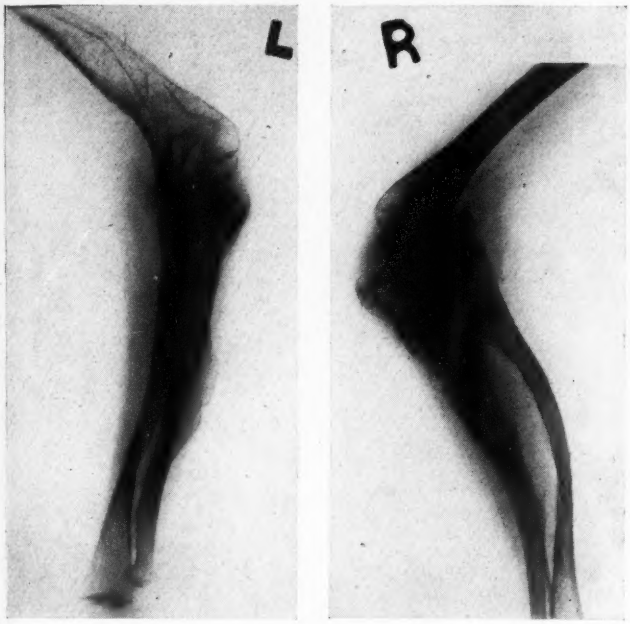

FIG. 5.—*Case* 2. Left and right arms (Jan., 1942) showing great reduction of callus around lower third of left humerus, and bony excrescences on left ulna. Right arm shows marked irregularity of surface of ulna, with a large bony outgrowth, of low density, projecting forwards and laterally, and, in addition, dislocation of head of radius.

FIG. 6.—*Case* 2. Right forearm (July, 1947) showing thickening of the ulna still present, though less than it was a year previously, when it was causing pressure-absorption of the radius.

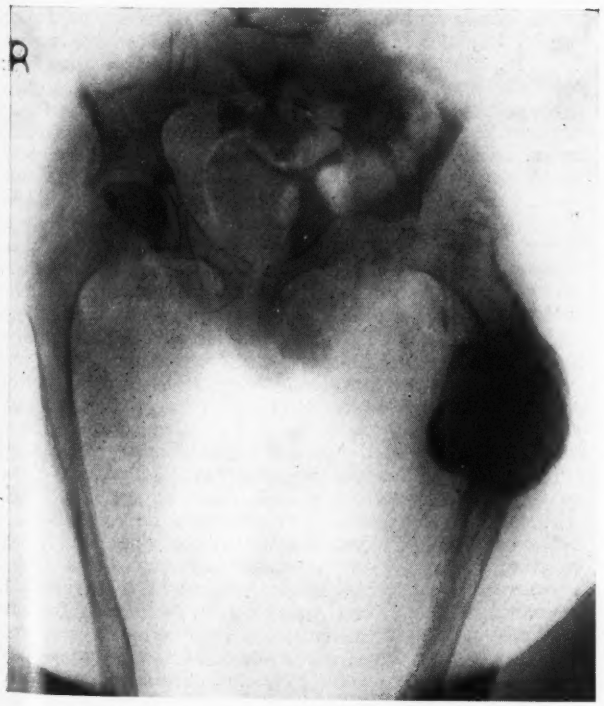

FIG. 7.—*Case* 2. Pelvis and femora (Jan., 1942) showing fracture of left femur with excess of callus forming a rounded mass of unusual density, with less dense extensions along the shaft of the bone.

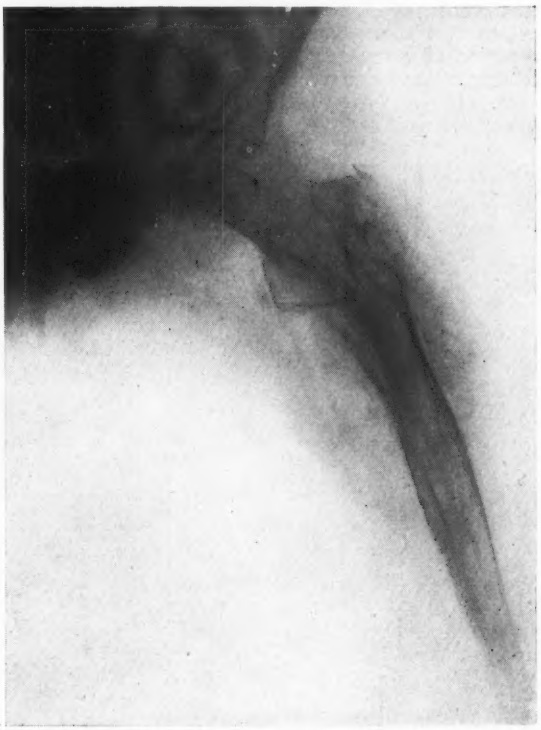

FIG. 8.—*Case* 2. Left femur (June, 1942) showing hyperplastic callus four months after osteotomy for correction of deformity. Callus is much less dense, but is still excessive and widely distributed along the shaft.

of the excess of bone on the ulna had been absorbed (*Fig.* 6). Pelvis is now indented. Excess of callus still present on both femora, with several bony excrescences on these bones and on the pelvis (*Figs.* 9, 10). (For later films and history I am indebted to Mr. J. S. Batchelor.)

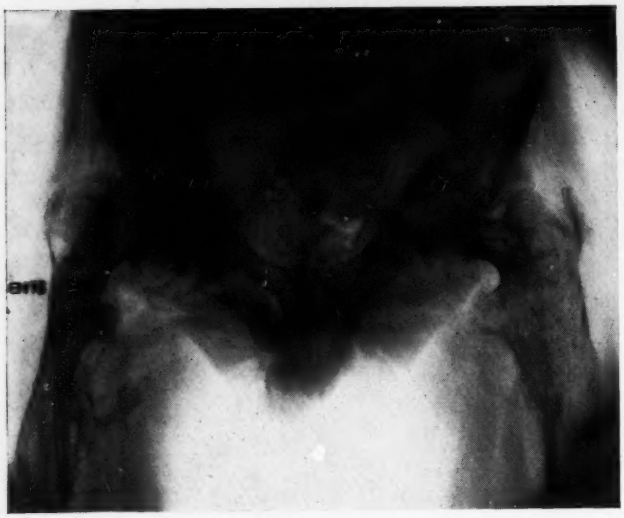

FIG. 9.—*Case* 2. Pelvis and upper femora (July, 1947) showing their condition at the age of 13. In addition to some excess of callus still present, there are bony excrescences from the ilia as well as the femora. The femoral heads have slipped: the femoral necks are unduly small.

Comment.—An undoubted case of osteogenesis imperfecta. The generalized osteoporosis increased with age and prolonged recumbency up to 10 years. Early dense callus followed fracture of both femora: in the left thigh diffuse hyperplastic callus developed later. Excessive callus formed around a fractured humerus, but this was eventually absorbed. Bony enlargement of one ulna occurred without a provocative fracture, increased over a period of years, and finally showed signs of absorption. Multiple bony excrescences were a marked feature of the case. Curious mottling has recently developed in the lower femoral and both tibial epiphyses.

FIG. 11.—*Case* 3. Femora (April, 1936) at age of 14 months, showing large masses of callus around both femoral shafts. That around left femur, almost certainly fractured, is of the butterfly shape referred to in the text; it varies in density, but is generally denser than normal early callus.

FIG. 10.—*Case* 2. Femora (July, 1947) showing the remains of the excessive callus on the shafts. The irregular density of the enlarged condyles is peculiar.

Case 3.—E. R., female. Normal at birth. Swelling of right shoulder at 7 months, soon followed by swelling of left shoulder and lower ends of both femora. Periosteal shadows present around shafts of several of the major long bones at 1 year. X-ray examination at 14 months showed enormous masses of callus of varying density around both femora (*Fig.* 11). On the right the callus involved the whole shaft; on the left it was of the butterfly type. Enlargement of upper third of left humerus: the excess was mostly absorbed later. At 19 months marked thickening of both thighs. Could crawl but not walk. Wassermann of mother negative. At 20 months a fall injuring left leg produced more callus. At nearly 3 years, lower two-thirds of both femoral shafts were enlarged to about three times the normal. Right leg half an inch longer than left. Fall on right elbow followed by a large bony thickening of lower part of humerus, with some limitation of extension of the joint. Mother stated great swelling invariably occurred if the child knocked herself. One attack of nose-bleeding. Greenstick fracture of right radius at 3½ was followed by gross thickening. Curvature of left femur increased: was dealt with by osteotomy later. Dr. Brailsford published radiographs of femora at 4 (1943). At 5, films showed enlargement, with cancellous bone, of most of shafts of both femora still present, with many bony excrescences (*Fig.* 12). At the osteotomy of the left femur "the periosteum and surface of the bone appeared normal. The cortex was egg-shell in thickness; the medulla soft and fatty." Operation followed by a flare and great swelling (*Fig.* 13). A mass of intensely dense callus appeared on inner side of lower end of left humerus, preceded by a few days' pain, but no definite injury and certainly no fracture: petechial hæmorrhages appeared in the overlying skin. Mass was tender, and covered by slightly hyperæmic skin. Flexion of elbow impossible above right angle (*Fig.* 14). Child rather small. General osteoporosis of whole skeleton. Vertebral bodies biconvex, and very translucent. Radii and ulnæ showed several bony excrescences on their interosseous borders.

FIG. 12.—*Case* 3. Both femora (March, 1940) at age of 5 years, showing the gross enlargement of the greater part of both shafts, with many bony excrescences. Note an outgrowth from the right ilium, and possibly another from the left, and the increased density in the region of the epiphysial lines near the knee-joint as frequently seen in osteogenesis imperfecta.

Fig. 13.

FIG. 13.—*Case* 3. Left femur (May, 1940) two months after osteotomy for anterior bowing, showing intensely dense, and apparently structureless, callus on the outer side of the shaft.

Fig. 14.

FIG. 14.—*Case* 3. Left humerus, showing dense callus on anterior aspect of lower end, and sessile bony outgrowth from middle of shaft. No definite injury. No fracture.

Blood examination negative. Wassermann negative, mother and child. No improvement followed treatment on special vitamin diet.

(Under care of Mr. G. F. Allan.)

Comment.—Apparently a definite case of osteogenesis imperfecta displaying all the special features discussed in this article—intensely dense callus after injury, both with and without fracture, enormous amount of hyperplastic callus around both femora, and multiple bony excrescences on many bones.

Case 4.—J. M., aged 15. Small thin boy. Typical fragilitas ossium with X-ray evidence of innumerable healed fractures. History of at least 38 previous fractures. Sustained a supracondylar fracture of left femur on Jan. 10, 1944, when getting off bed-pan. Two weeks later developed œdema of ankle, foot, and knee. X-ray examination at six weeks showed fracture united by abundant callus (*Fig.* 15). Marked œdema of leg developed with dilated veins over the knee and effusion into joint. Thigh hot, swollen and firm. Sarcoma suspected, and biopsy performed eight weeks after fracture. The incision revealed on division of muscles, a greyish mass, a wedge of which was removed down to the bone. Sections were reported as hypertrophic fracture callus. Later X-rays showed a steady increase in the callus shadow till the swelling involved the whole thigh (*Fig.* 16). A further fracture in the trochanteric region, surrounded by dense nodular callus, as was the fracture in the lower third, and a slipped epiphysis of the femoral head, were discovered. X-ray taken 24 weeks after fracture showed callus extending whole length of bone. A month later the swelling was diminishing and by the fortieth week it had considerably diminished and the œdema and dilated veins had disappeared: an X-ray then showed trabeculated porous callus extending the whole length of femur. This callus was still present fifteen months after injury (*Fig.* 17). During this period

FIG. 15.—*Case* 4. (Feb. 22, 1944.) Left femur 6 weeks after fracture, showing fragile appearance of bone and large mass of callus around fracture site. (Biopsy 2 weeks later.)

FIG. 16.—*Case* 4. (April 21.) Left femur 8 weeks later than *Fig.* 15, showing great extension of the callus, which is unusually dense.

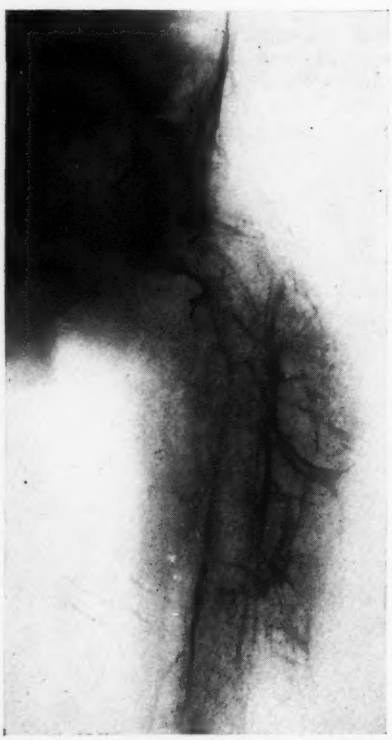

FIG. 17.—*Case* 4. (April 18, 1945.) Left femur a year later, showing callus now porotic, but still present in abundance, and extending to the femoral neck. An old fracture is seen in the upper third of the shaft.

other fractures of left leg developed—upper end of tibia and fibula and Pott's fracture: no excessive callus formed around these. About three months after the left femur had fractured a supracondylar fracture of right femur occurred and an almost identical mass of callus developed in relation to it. The mass of callus extended up its whole length as far as the neck, just as on the left side (*Fig.* 18). Fractures in the upper limb bones had

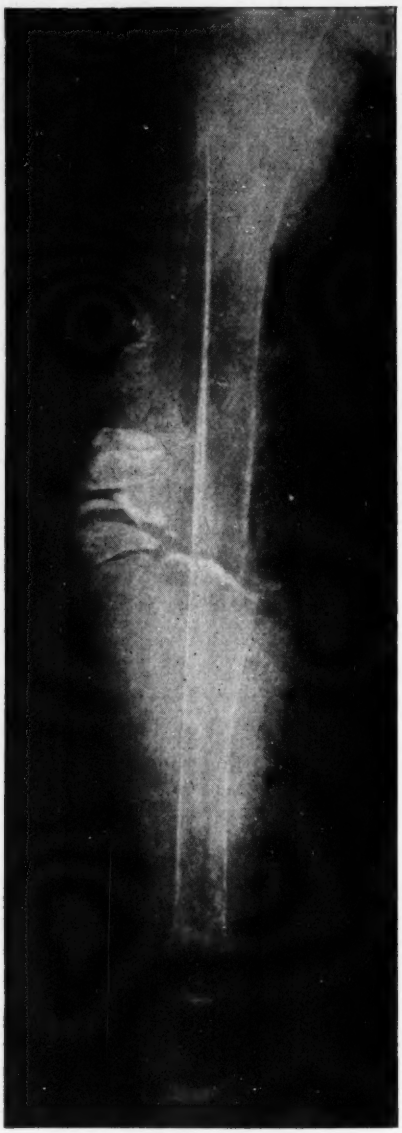

FIG. 18.—*Case* 4. (Oct. 18, 1944.) Right femur 26 weeks after fracture, showing enormous mass of callus filling the thigh and extending up to the neck of the femur. The site of the supracondylar fracture can just be detected. There is also a fracture through (or gap in) the callus opposite the middle of the femur, but no signs of disturbance of the continuity of the bone itself.

resulted in deformity and limited movement of the elbows. Head of the radius was dislocated backwards in both arms. Limited movement in both hands and both hips. Muscular development extremely poor. Blood examination revealed nothing of importance.

(Under Professor Sir Harry Platt.)

Comment.—A severe case of osteogenesis imperfecta in which a supracondylar fracture of one femur resulted in an excessive amount of callus and such enormous swelling of the limb that sarcoma was seriously considered.

A biopsy was performed and a malignant neoplasm excluded (*see* Professor Baker's report). A similar mass of callus developed in the opposite thigh following an almost spontaneous fracture of the femur.*

Case 5.—B. G., female. No deformities until she was 3 years old, when following whooping-cough, measles, and pneumonia, deformity of legs, with difficulty in walking, was present. At 7 years her head was large, with a mop of black straight hair and somewhat Mongolian facies. General obesity. Muscles hypotonic. Shuffling, waddling gait. Outward bowing of humeri. Marked anterolateral bowing of tibiæ. Blood examination revealed nothing of importance. Wassermann negative.

FIG. 19.—*Case* 5. Pelvis and femora at age of 8 years, showing the enlargement of the shafts of the femora and the texture of these bones.

X-ray Examination.—Humeri thicker than normal, with irregular outline and density: shafts honeycombed in places; cortex thin. Old fracture of left ulna with imperfect union and some angulation. Phalanges of hands poorly calcified. Lower two-thirds of both femoral shafts show considerable enlargement, which ends above rather abruptly on the right side. Bone forming the enlarged portions is porous: appearances are consistent with possible old fracture (*Fig.* 19). Bilateral coxa vara. Both tibiæ and fibulæ show marked curvature in their lower halves. Carpal and tarsal bones present somewhat dense centres, some in the form of a ring, with less dense bone around. Nothing noteworthy in spine. When examined at 8½ years she was definitely dwarfed and very fat, especially the abdomen. No enlargement of liver or spleen. Sclerotics not blue. Few teeth remained, but these were well calcified. Many scars on forehead, nose, lip, and cheek, and there was no septum between the nostrils. No hare lip. Deformities of the bones as described above still present.

(Under Mr. L. A. Key.)

Comment.—Though the films of some of the bones are suggestive of old fractures, and there is a definite fracture of one ulna, this case is not regarded as one of osteogenesis imperfecta. It is included in the series, with some hesitation, because the condition of the femora is so suggestive of that seen in the other cases, and is quite unlike any other type of case with symmetrical enlargement of the femoral shafts with which we are familiar.

Case 6.—E. H., female. Fractured left ankle at 9 years. At 10 years pain in left leg above knee. Admitted to hospital three weeks after onset, when biopsy was done. Case was fully reported by Dr. Gwendoline Hilton in 1934 as one of familial chondrodystrophy with rheostosis. At that time, without any signs of fracture, there was a large bony mass connected with left femur, which was treated by X rays. Further increase in size of the mass took place after the first course, but improvement followed further treatment. There was similar hyperostosis of right femur, and bony excrescences on the scapula, left fibula, and other bones, with ridges on some metacarpals and metatarsals. Full investigation of blood, etc., revealed nothing of interest. Biopsy of left femur showed " Bony trabeculæ having no definite arrangement. Ramifying between these are stout columns of very vascular and very loose connective tissue. Along the line of separation between the fibrous tissue and bone are a number of osteoclasts. From the appearance of the neighbouring bone little absorption is taking place ". Height was 3 ft. 6 in. Epiphysial lines of all the bones were thickened and wavy. There was bilateral dislocation of the radial heads. The girl's mother was small and had a hyperostotic ridge on one femur, and dislocation of the head of the radius in at least one arm. One maternal aunt was said to be like the mother, while another, aged 24, was severely deformed and showed bony changes similar to the patient, but worse. Some of her joints were affected, e.g., the elbows, these being overshadowed by irregular confluent bony shadows in radiographs.

When seen at age of 23, patient had been admitted to hospital after a fall during a cerebral ' black-out '. She said she often got these ' black-outs ', which lasted a few minutes. Complained of pain in left hip and inability to move herself. Right leg was short: a special boot was worn. Movements of both hips much impaired, especially left, and limitation of elbows, of pronation and supination and of flexion of the wrists. Blood examination, nothing of importance. In addition to family history quoted above, a male cousin was said to have ' deformed arms '.

X-ray Examination.—Skull thin. Ribs unusually horizontal, chest being broad. Fusiform enlargement in upper third of right humerus. Bony excrescences on radius and ulna. Pelvis showed bilateral protrusio acetabuli, with fracture of left acetabular floor, displacement being minimal. Pubes and ischia were curiously shaped. Both femora rather translucent, with additional ensheathing bone along both shafts, inside the right and both inside and outside the left. Sharp spikes of bone projected downwards from lower margins of great trochanters. Bony ridges and excrescences on posterior aspect of upper thirds of tibiæ and on most of shafts of both fibulæ. At 25 (1946) the hyperostosis showed little change and was still widely distributed (*Fig.* 20). Bony outgrowths from both anterior superior iliac spines were seen, and some subperiosteal new bone above right acetabulum. Each ilium gave the appearance—almost certainly a false appearance—of being occupied by a

* Another large mass of callus recently formed spontaneously about the left femur. No definite fracture. Œdema extended to left foot and beyond the limits of the limb above. Swelling now subsiding—four months from onset.

large cyst. Both humeri showed bony thickening in the upper halves of their shafts, which were otherwise slender and curved. Head of radius in both arms grossly abnormal in shape. Genu valgum present. Fibulæ slender and curly, with excrescences the whole length of shafts (*Fig.* 21). Scoliosis present: transverse processes and tri-radiate. Acetabulæ very deep. Irregular bony outgrowths on inner side of upper and middle thirds of shaft of right femur. Large mass of irregular bone around the lower half of right humerus, especially just above external epicondyle, and extending more than halfway up the shaft: no sign of recent fracture. Irregular

FIG. 20.—*Case* 6, at age of 25 years. Pelvis and femora showing the cyst-like appearance of the ilia, the abnormal shape of the ischia and pubes, the protrusio acetabuli, with fracture in the left acetabulum, and the various permanent (?) bony thickenings and excrescences on the femora.

FIG. 21.—*Case* 6, at age of 25. Tibiæ and fibulæ showing the deformities and the widespread bony excrescences on the latter bones.

of fifth lumbar unusually long. The excrescences from the right radius and ulna seemed to have fused. Skull thicker than it was two years previously: pituitary fossa appeared to be closed in.

(Under Mr. V. C. Snell.)

Comment.—Not a case of osteogenesis imperfecta. When first observed by Dr. Hilton the bony outgrowths on the femora were similar to those seen in other cases in this series (*see* radiographs published in 1934). The recent films, though displaying unique features, also show bony thickenings and excrescences typical of the condition discussed in this paper.

Case 7.—I. J., male, aged 6. Only child. Well till 3 months old, when lost use of right arm and leg for two months. Had broken practically every bone at different times. Within last month broke left arm. Cannot walk. Growth very stunted. Curvature of both femora, left tibia, and both humeri, and to a less degree of radii and ulnæ. Face thin and peaky. Pigeon breast. Sclerotics not unduly blue. Blood examination revealed nothing noteworthy.

X-ray Examination.—General osteoporosis. Vertebral bodies shallow. Ribs horizontal. Pelvis indented density above and below epiphysial lines of femur and tibia adjacent to the knee-joint. Skull thin.

(Under Dr. W. G. Wyllie and Mr. Denis Browne.)

Comment.—An undoubted case of osteogenesis imperfecta showing some tendency to excessive callus formation and permanent bony excrescences. (Prints of radiographs not suitable for reproduction.)

Case 8.—A. E., aged 4½. Thin, poorly developed boy showing in X-rays thin fragile bones with innumerable healed fractures. Nothing noticed wrong with child in first twelve months, but between ages of 1 and 4½ years he sustained at least ten fractures, with little or no violence. In 1941, when about 18 months old, sustained spiral fracture of midshaft of right femur. X-rays showed oblique fracture and nothing unusual in the cortex. Mass developed in relation to the fracture; within four weeks this had reached considerable size, and in the X-ray central part around the fracture site showed much calcification, and a layer of periosteal thickening extending up the bone (*Fig.* 22). Thigh was then greatly enlarged, hot and showed many distended veins: closely resembled a rapidly growing sarcoma. Biopsy was performed five weeks after fracture. Disarticulation at hip considered, but regarded as too risky and of doubtful value. X-ray treatment preferred.

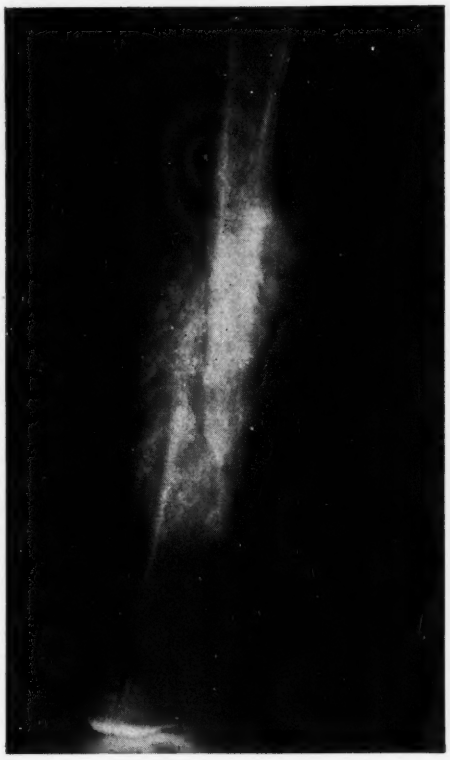

FIG. 22.—*Case* 8 (Aug. 11, 1941). Right femur four weeks after fracture, showing callus developing round fracture site. (Biopsy one week later.)

FIG. 23.—*Case* 8 (Nov. 24). Right femur nineteen weeks after fracture, showing large calcified mass filling the thigh.

Note made seven weeks after fracture includes "Huge swelling of right thigh from below knee to above groin, œdematous, hot, distended veins on surface. Tumour fungating through biopsy scar". Mass in thigh gradually became harder and biopsy incision healed: œdema disappeared and mass became more compact. An X-ray taken four months after injury (*Fig.* 23) shows large calcified mass filling whole thigh: nodules of this could be felt as hard subcutaneous masses. The fracture had united. Thigh continued to diminish in size, and patient was eventually able to walk. An X-ray taken nearly three years after fracture (*Fig.* 24) shows callus

FIG. 24.—*Case* 8 (June 7, 1944). Right femur nearly three years after fracture, showing fusiform mass of reconstructed porosed bone occupying the middle half of the femoral shaft.

mass had been largely reconstructed and now forms part of an expanded but porosed and trabeculated femoral shaft, with no signs of fracture site.

(Under care of Dr. Edith Paterson.)

Comment.—A case of osteogenesis imperfecta in whom an oblique fracture of one femoral shaft was followed by the development of such an excessive amount of callus, with great swelling of the limb and distended veins, as to suggest sarcoma and call for biopsy (*see* Professor Baker's report). The 'tumour' fungated through the biopsy wound, was treated with X rays, and then gradually subsided.

HISTOLOGY OF HYPERPLASTIC CALLUS

(Professor S. L. Baker)

Material.—Biopsies were performed in *Cases* 4 and 8 at a comparatively early stage in the development of the callus and the following account is based on this material.

In *Case* 4 the biopsy was taken eight weeks after fracture and two weeks after the radiograph shown in *Fig.* 15. In *Case* 8 the biopsy was taken five weeks after fracture and one week after the radiograph shown in *Fig.* 22. In both cases only a small part of the biopsy showed calcified tissue corresponding to the outer margin of the X-ray shadow; the bulk of the material corresponded to the rapidly growing outer part of the callus not yet calcified and forming a soft tissue swelling invisible radiographically. The tissue differed strikingly from 'normal' fracture callus in appearance. Normal callus would at an earlier stage than this (by about 3 weeks) have formed a firm fibrous mass containing a spongework of calcified and calcifying bone trabeculæ. Here the bulk of the tissue was gelatinous and semicartilaginous, resembling to the naked eye a myxochondroma or a chondrosarcoma; only in its deeper part did it show reddish, firm, partly calcified tissue, not unlike normal fracture callus. The amount of tissue removed from *Case* 8 was abundant and consisted mainly of the gelatinous tissue. In *Case* 4 it was less abundant and consisted of a wedge extending from the surface down into the calcified zone seen in the radiograph (*see Fig.* 15).

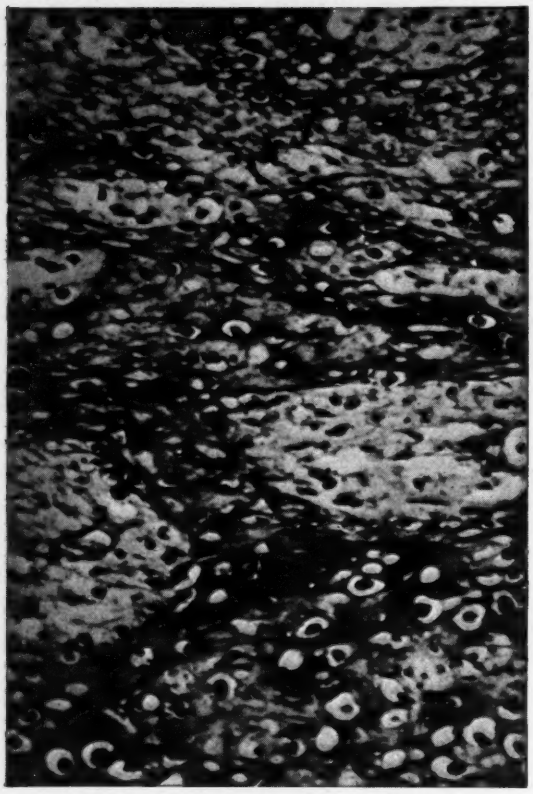

FIG. 25.—*Case* 8. Chondroid tissue (Heidenhain's Azan) showing strands of fibromucoid tissue with perivascular reticular tissue (lighter areas). (× 165.)

Microscopical Features.—The following is a summary of the main microscopical features, described in more detail in a previous paper (Baker, 1946). Microscopically the tissue from both these cases showed essentially the same histological features, the bulk of the material consisting of a fibromucoid, cartilage-like tissue (*Fig.* 25). Histologists (Schaffer, 1930) have used the term 'chondroid tissue' for similar semicartilaginous tissue, and this seems the best term to apply to transitions between fibrous, mucoid, and cartilaginous tissue met with under pathological conditions.

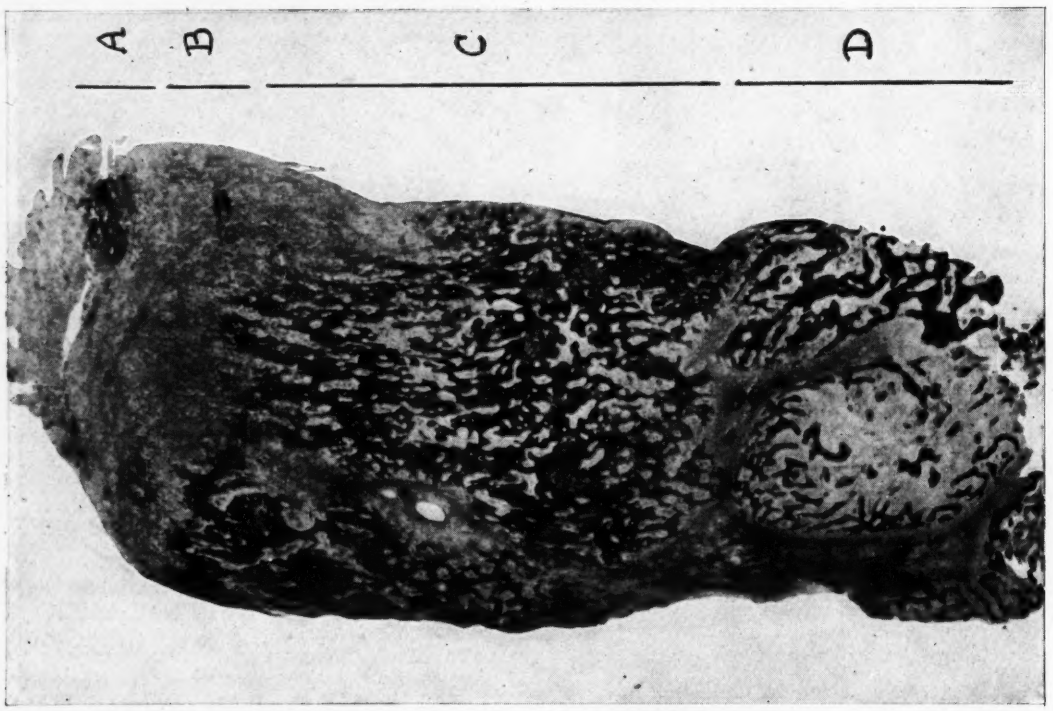

FIG. 26.—*Case* 4. Section of callus showing the various zones described in the text. Zone D, consisting of calcified trabeculæ, is comparable with the normal callus seen in *Fig.* 27. Stain: Weigert's iron hæmatoxylin and van Giesen. (× 8.)

The chondroid tissue in this material showed transitional forms grading into true cartilage in some places and into woven bone in others.

By selecting suitable pieces of tissue it was possible to observe the various stages in the development of the chondroid tissue and also transitions from chondroid to true cartilage or to woven bone.* *Fig.* 26 shows a general view (× 8) of a section of a piece of tissue from *Case* 4 which passed from the superficial to the deep part of the biopsy specimen. It consists mainly of chondroid tissue permeated by irregular vascular (marrow) spaces (pale channels in *Fig.* 26). Various zones can be seen; externally (Zone A) is the advancing edge of the tissue where it is replacing the muscle and connective tissues of the thigh; Zone B shows the early stages of chondroid formation; Zone C is fully developed chondroid tissue with islands of true cartilage and also transitions to woven bone; Zone D shows trabeculæ composed either of calcified woven bone or of calcified cartilage. To the naked eye Zones A, B, and C correspond to the pale mucinous semicartilaginous tissue mentioned above, while Zone D is the reddish firmer tissue.

The tissue from *Case* 8 was less regularly arranged than the piece from *Case* 4 illustrated here and it contained larger amounts of the gelatinous early chondroid (Zone B), but the various zones seen could all be recognized in it and material from either case is used to illustrate the various appearances described below.

Zone A. Œdema with Destruction of Muscle.—This consists of œdematous tissue separating collections of atrophic muscle-fibres and containing partly absorbed fragments of collagen fibre.

Zone B. Mucoid Œdema, Cellular Proliferation, and Early Chondroid Formation.—This is œdematous and mucoid tissue containing numerous young connective-tissue cells so that many areas resemble a tissue culture of fibroblasts (*Fig.* 28). The earliest development of chondroid is seen in this zone in the form of collections of more rounded chondroblastic cells (*Fig.* 29) around which develops mucoid tissue containing a network of collagen fibres. Another feature of this zone is a marked perivascular œdema around many of the newly formed thin-walled vessels.

Zone C. Chondroid Tissue.—This is composed mainly of chondroid tissue permeated by vascular channels which divide it up into irregular masses and strands.

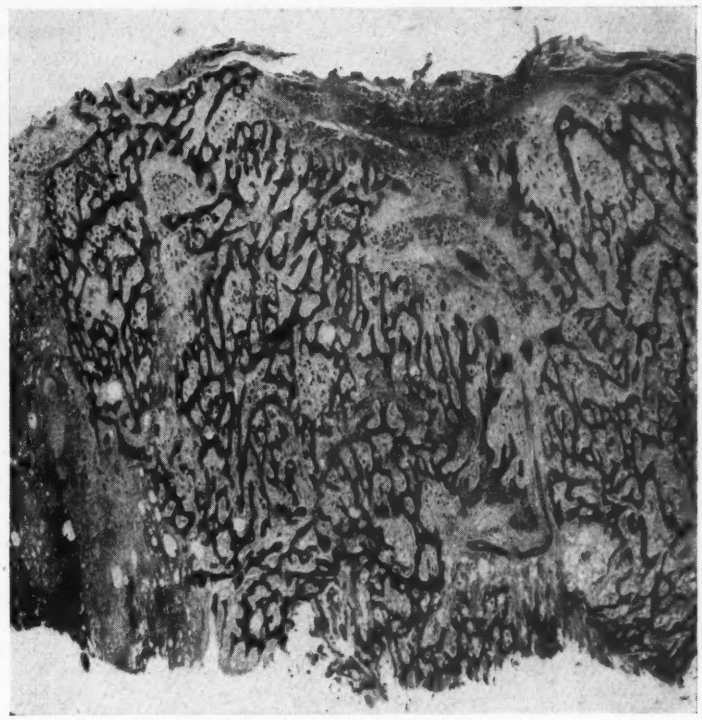

FIG. 27.—Normal callus for comparison with *Fig.* 26. Three-weeks-old callus from a fractured femur; it is composed of trabeculæ of calcified woven bone lying in fibrous tissue. This is bounded externally (above) by a layer of muscle. It is comparable to Zone D in *Fig.* 26; there is no tissue corresponding to Zones A, B, and C. Stain: Mallory. (× 8.)

It also contains islands and strands of true cartilage. This chondroid tissue may be described briefly as a fibromucoid tissue, intermediate in structure between cartilage and woven bone; all three tissues —cartilage, chondroid, and woven bone—occur in this zone, and 'chondrosteoid' tissue intermediate between chondroid and woven bone is seen. Patchy calcification occurs in this zone, mainly in the centre of the islands of cartilage and in the strands of woven bone; the chondroid shows for the most part little or no calcification.

Zone D. Calcified Trabeculæ.—This is composed of trabeculæ formed of calcified woven bone, but also of calcified cartilage, the whole forming an irregular cancellous tissue with marrow spaces containing fine reticular connective-tissue and thin-walled vessels.

Scattered osteoclasts are seen on the surfaces of both calcified and non-calcified tissue in Zone C and of the calcified trabeculæ in Zone D.

DEVELOPMENT OF THE CALLUS

Sections such as that in *Fig.* 26, extending in from the growing edge of the mass, show a picture which can be interpreted as an orderly sequence of

* The term 'woven bone' is not in general use in English and therefore needs an explanatory note. It refers to the non-lamellar bone commonly found under all sorts of pathological conditions where new bone is being rapidly formed, e.g., provisional callus of a fracture. There is no standard term for it in English; it has been called 'fibre-bone', 'coarse-fibred bone', 'immature bone', etc. Ebner (1875) separated this type from lamellar bone and called it 'geflechtartiger Knochen'; a term commonly used in the German literature. 'Woven bone' is an English translation of this term and is used by H. M. Turnbull. It is a good term because it indicates the essential difference between the irregular woven network of fibres forming the matrix of this type of bone in contrast to the regular layered arrangement of fibres in lamellar bone.

events, the most recent being at the surface where changes are occurring in the muscle and connective tissue leading to their dissolution, and the oldest is seen in the deepest part where a calcified cancellous structure has replaced the normal tissues. The appearances suggest that the hyperplastic callus in these two cases passed through the following stages :—

1. An œdema of the muscle and connective tissue similar to an inflammatory œdema and presumably determined by changes in the vascular condition of the region.

2. Atrophy of the muscle and connective tissue, in place of which develops,

3. An œdematous mucoid tissue containing numerous cells of fibroblast type. These cells differentiate in various directions ; many of them produce bundles, brushes, and webs of collagen fibrils ; some assume a rounded shape (chondroblasts)

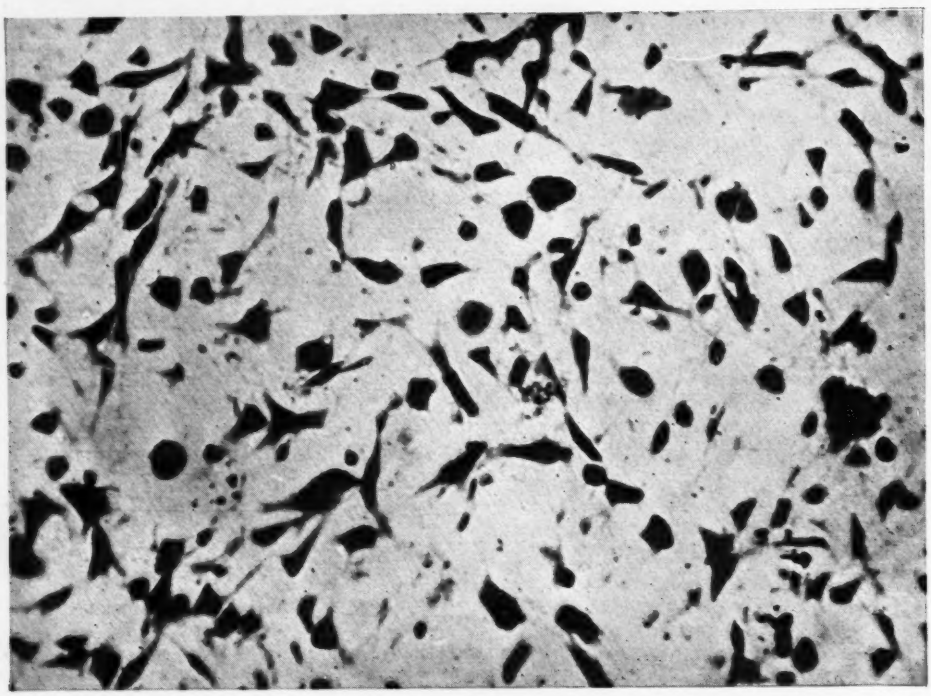

FIG. 28.—Zone B, *Fig.* 26. Frozen section (hæmatoxylin and Sudan IV), showing proliferation of fibroblasts. (× 335.)

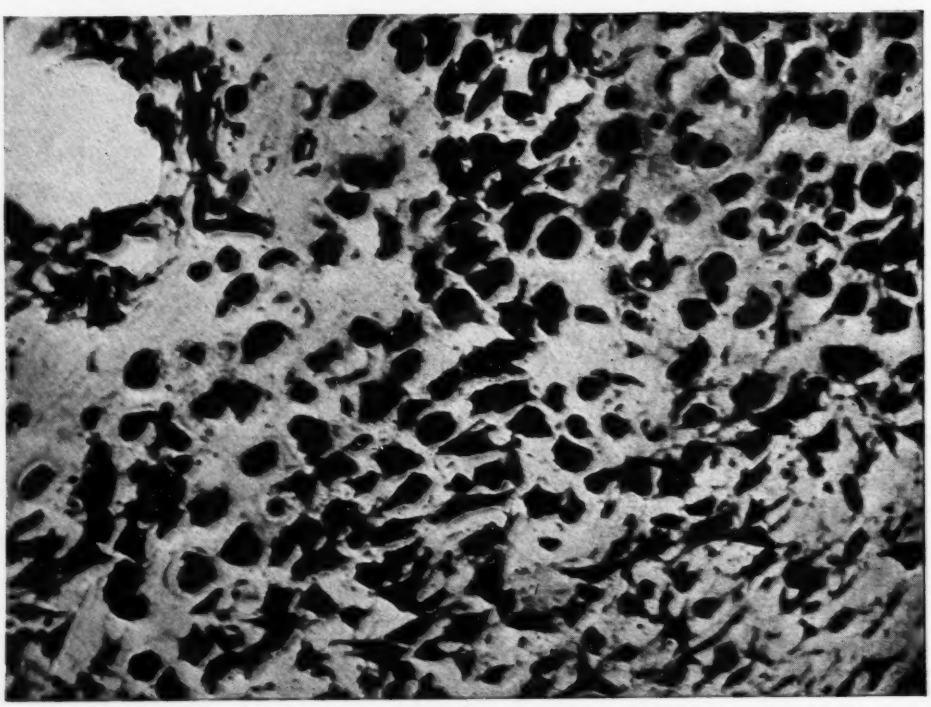

FIG. 29.—The same section as *Fig.* 28, showing a collection of more rounded (chondroblastic) cells. (× 335.)

and become surrounded by increasing amounts of mucoid tissue, thus forming a fibromucoid (chondroid) tissue or, with an increased development of basophil mucoid which masks the fibres, true cartilage. Other cells remain stellate in shape, with long cytoplasmic processes, and become included in the fibromucoid; these develop into the bone cells in the chondrosteoid tissue, which, with increasing amounts of collagen, assumes the characters of woven bone. In the zones of œdema immediately around vessels neither collagen nor mucoid develops, but a fine reticulin network associated with scattered stellate cells; this forms the earliest stage of the perivascular (marrow) space.

The calcified cancellous tissue of Zone D forms the outer part of the X-ray shadow. It develops by a remodelling and further calcification of the deeper tissues in Zone C. The remodelling is carried out by osteoclasts seen, as noted above, in Zones C and D. An irregular network of calcified trabeculæ is formed. In some areas these are fine trabeculæ of woven bone; in others (*Fig.* 26, right hand side of Zone D) coarse trabeculæ and irregular masses of calcified cartilage.

Comparison of Normal and Hyperplastic Callus.—Comparing normal with hyperplastic callus we find that normal callus (*Fig.* 27) is formed of a network of trabeculæ of woven bone lying in a fairly dense connective tissue. This is bounded externally (top of *Fig.* 27) by the surrounding intact muscle. The callus has invaded the muscle and remains of atrophied muscle (black dots in between trabeculæ in *Fig.* 27) are found in the fibrous tissue. There is no zone of œdematous mucoid or chondroid tissue (*Fig.* 26, A, B, and C).

Relation of Histology to Clinical and Radiographic Features.—The sarcoma-like clinical appearance of the active stage of this hyperplastic callus is well explained by the mass of soft vascular rapidly-growing tissue seen in this material. Only a little calcified tissue, corresponding to the outer parts of the X-ray shadow, is included in these biopsies. This has the general characters of fracture callus and would become denser in the deeper (older) part by continual deposition of new bone on the surface of these trabeculæ.

The very dense radiographic shadow seen in many cases at the comparatively early stage in the development of the hyperplastic callus needs explanation, but unfortunately these biopsies do not include the denser parts of the callus. One never finds calcification of tissue other than bone or cartilage in normal fracture callus, nor is there any sign of such calcification of the soft tissues in this material. The most probable explanation of the very dense radiographic shadows is therefore the presence of considerable amounts of calcified cartilage forming irregular thick trabeculæ and small masses such as are seen in the right-hand side of Zone D in *Fig.* 26. *Fig.* 30 shows such masses of calcified cartilage in another area in the deeper part of the biopsy specimen from *Case* 4. This is vesicular-celled cartilage, islands of which are common in normal callus. It is similar to the vesicular-celled cartilage at the normal epiphysial line of a growing bone, and undergoes the same fate, being first calcified and then eroded and replaced by bone. It is clear that there is an excessive amount of cartilage in this hyperplastic callus; an excess of calcified cartilage is therefore to be expected in the intermediate stage of the process. This would account for the early development of a dense radiographic shadow, subsequently replaced by a less dense shadow as the calcified cartilage is eroded and replaced by less massive bone trabeculæ. It is clear from the later radiographs that the callus mass may eventually become greatly porosed, undergoing the same change as the rest of the skeleton.

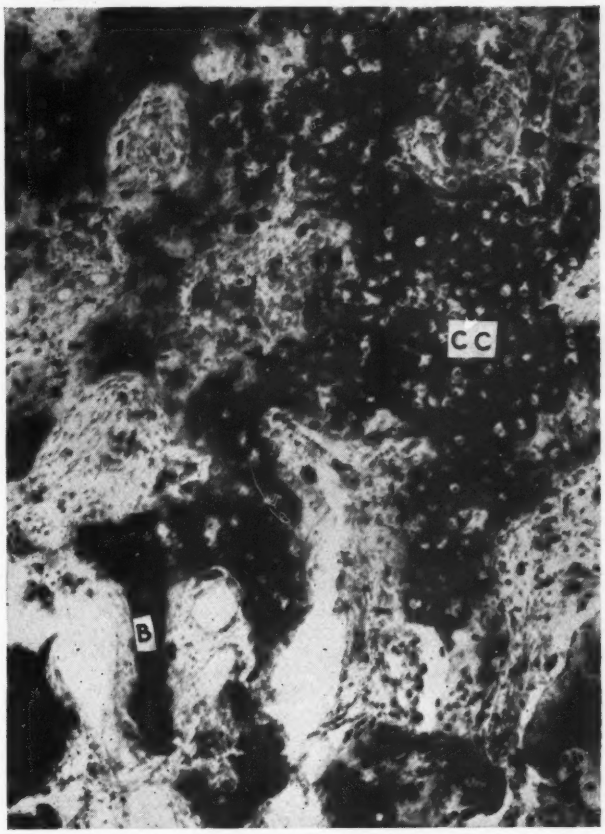

FIG. 30.—An irregular mass of calcified vesicular-celled cartilage (C C) connected by an irregular trabecula with a small trabecula of woven bone (B). Undecalcified frozen section; stain, hæmatoxylin. (× 70.)

DISCUSSION

It is clear that there is a definite relationship between excessive callus production and osteogenesis imperfecta. This excessive reaction may follow on a fracture or may occur with the bone apparently intact. It is limited to certain sites—most commonly the femur—and has developed on femur fractures while simultaneous fractures at other sites healed without excessive callus.

We cannot agree with Brailsford's suggestion that scurvy plays a part in this reaction. In Cases 4 and 8, here described, the sequence of events has been followed closely from the initial fractures to the final reconstruction of the bone; there was no evidence of scurvy or other hæmorrhagic disease in these cases. In cases without definite evidence of fracture the evidence for scurvy is also lacking. The

shadows cast by the exuberant callus are quite unlike the smooth shadows resulting from subperiosteal hæmorrhages in scurvy. Moreover, in scurvy, as Brailsford admits, these shadows disappear completely under treatment and leave no permanent disfigurement of the bone. Though two cases show some excess of central calcification on either side of the epiphysial lines of the femur and tibia at the knee, an appearance not infrequently seen in osteogenesis imperfecta, this is quite unlike the patches of intense calcification, tending to be triangular in shape and limited by the epiphysial line, that have been seen in the metaphyses and adjacent parts of the shafts in adolescent scurvy. Nor are these latter appearances seen in the older cases in our series (*see* Cassidy, 1923, and Pugh, 1925). With one exception already referred to, none of the cases has exhibited hæmorrhages in the skin, the gums, or the urine. Also the evidence is strongly against there being a deficiency of vitamin C in any of the cases.

The excessive callus cannot be attributed to excessive local hæmorrhage and muscle damage, of which there is no evidence. In *Case* 4, for instance, where massive callus developed in two successive fractures, one of the left and the other of the right femur, the trauma was in both instances minimal and the biopsy material (from the first fracture) showed no signs of collections of blood-pigment or other evidence of extensive hæmorrhage. Moreover in traumatic myositis ossificans complicating a fracture the callus is rarely extensive and then only in a case that has been handled unwisely, and it never extends along the shaft of a bone to the degree seen in the cases here reported. It is confined to the single bone damaged, and if treated by prompt and efficient immobilization, in children at any rate, it usually disappears completely.

Cases of doubtful origin and characterized by thickening, often symmetrical, of the major long bones have been published from time to time, but these differ in important details from those now under discussion. (*See* Engelmann, 1929; Bakwin and Krida, 1937; Riley and Schwachman, 1943; Hirsch, 1944.)

Apart from trauma it is difficult to assign any exciting cause for the excessive reaction. It is curious that in spite of the poor osteoblastic function in osteogenesis imperfecta, the local reaction to the stimulus of a fracture is good. Until we know more about the general and local mechanisms controlling osteoblastic activity, it is impossible to explain either the good callus formation usually shown in these cases or the occasional excessive reactions here reported.

We are greatly indebted to the colleagues and friends who have generously supplied us with clinical data of the cases under their care.

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SOLITARY PLASMOCYTOMA OF BONE WITH RENAL CHANGES

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SOLITARY tumours of bone-marrow composed entirely of plasma cells are now well recognized but are exceedingly rare. Willis (1941), in an excellent review of the literature, was able to find only 12 cases adequately proven, whilst adding 1 more that he had observed himself.

The literature since that time contains a number of references to solitary plasma-cell tumours (myeloma) of bone, but when these are reviewed critically in the light of Willis's work, only 2 more cases can be allowed as being completely proved.

The first case is Case No. 1 of Gootnick's series (1945), where the tumour occurred in the right ilium of a 48-year old male. It was followed up for 3½ years without the appearance of Bence-Jones protein, or radiographic changes in other bones.

The second is the interesting case reported by Kaufman (1945) of a solitary tumour in the skull of a boy aged 14. This case was followed for four years. The weakness in this report is that no specific mention of complete radiographic study is made after the 4-year period. The author simply states: "the patient was perfectly well in every respect".

With these two cases and the one about to be reported the total number of solitary plasma-cell tumours of bone is brought to 16.

The association of kidney lesions and Bence-Jones proteinuria with multiple myelomatosis has been recorded on a number of occasions and specific abnormalities have been described. No record can be found, however, of the typical renal lesions occurring in association with a solitary tumour.

Willis (1941) has suggested various criteria to be observed in the diagnosis of solitary plasma-cell tumours of bone-marrow. First, the possibility that the tumour is merely the initial sign of generalized

myelomatosis must be excluded. Secondly, a histological diagnosis must be made. Radiographs of the entire skeleton, repeated at intervals, must be performed, and periodic testing of urine for Bence-Jones protein must be carried out.

A number of authors (notably Cutler, Buschke, and Cantril) have stressed the fact that plasma-cell tumours commencing in a solitary form subsequently metastasize to become multiple, with ensuing death. They suggest that it is only the rapidity of spread which produces the clinical picture of multiple myelomatosis. In other words, they assume a single disease process producing benign or solitary lesions capable in most instances of metastasis or malignancy.

As opposed to this reasoning, most pathologists believe that multiple myelomatosis is a multifocal systemic disease and that metastasis does not occur. Many believe that solitary plasmocytoma should be regarded as " a separate and distinct entity " (Willis, 1941).

It is considered of value to make a report of the following case, at the same time attempting to draw some conclusions from the findings of Bence-Jones proteinuria and renal tubular blockage in a case of solitary plasmocytoma of bone.

CASE REPORT

HISTORY.—A man (H. N.), aged 72, was admitted to the Westminster Hospital in November, 1946, complaining of severe pain at the lower end of the back and in both legs. The pain was worse on the right side.

He stated that he had been in good health until the beginning of February, 1946, when he developed pain in both hips and thighs, made worse by walking and bending. This pain slowly increased until May, 1946, when it was most severe at the " bottom of the spine " and a tender lump was noticed over the sacral region causing pain when weight was thrown on the right buttock.

In June, 1946, he noticed swelling of his ankles, which did not improve after rest in bed, and pain radiating down both legs. He was able to lead a reasonably active life, walking, and occasionally cycling, until the time of admission.

ON EXAMINATION (Nov. 29, 1946).—He was a fit-looking man, young for his years, intelligent and co-operative. The tongue was clean and moist. The only relevant finding was a large hard mass in the sacral region which extended over the whole width of the sacrum from the level of the upper border down to the coccyx. It was tender, and had ill-defined borders merging into the surrounding tissues. The centre of the tumour was elastic, with a suggestion of fluctuation.

On palpation per rectum the prostate was firm but only slightly enlarged. A large tender mass could be felt, evidently arising from the posterior wall of the pelvis and filling the pelvic cavity.

He had been constipated during the preceding three months, although he gave a history of regular bowel habits before that time.

He had some frequency of micturition at night (D/N=2/5), but he had no urgency or dysuria and did not have to strain.

The following investigations were performed :—

A punch biopsy was carried out by Mr. E. S. Lee on Dec. 19. Microscopically the specimen obtained showed a mass of cells of varying shape, about 60 per cent of which appeared to be typical plasma cells. A provisional diagnosis of myeloma, probably of plasma-cell type, was made.

In view of this discovery radiographs of every bone in the body were taken on Dec. 16, Jan. 9, 1947, and Jan. 16. No bony abnormality outside the sacrum was discovered.

The X-ray appearances of the sacrum showed a rapidly growing neoplasm of osteolytic type, involving practically the whole of the sacrum, and an adjacent area of both iliac bones, larger on the right than the left, with invasion across both sacro-iliac joints. The lumbar spine had descended slightly and the right transverse process of the 5th lumbar vertebra appeared to be involved (*Fig.* 31).

Urine was tested on numerous occasions and each time albumin and Bence-Jones protein were found. On Dec. 17, 1946, the total urinary protein was 235 mg. per 100 c.c.

Lactose-fermenting coliform organisms were grown from the urine on Dec. 2.

FIG. 31.—Radiograph of tumour three weeks before death.

Wassermann and Kahn reactions were both negative.

The blood-urea on Dec. 3 was 33 mg. per 100 c.c. and on the same day the serum-alkaline-phosphatase was 7 units whilst the acid-phosphatase was 2·1 units, all of which was alcohol stable.

The patient was discharged on Dec. 18, still in relatively good general health for readmission to the Irradiation Wards when a bed became available.

He returned to the hospital on Jan. 8, 1947, and his condition had changed considerably. He was no longer in robust health, but was drowsy and unco-operative.

He was constipated and his urine was loaded with albumin and Bence-Jones protein. He had no difficulty in passing water, the bladder was not enlarged, and there was no retention of urine. His urinary output was normal at 1,500 c.c. per day.

His condition deteriorated during the next weeks, and the blood-urea on Jan. 16 was 132 mg. per 100 c.c., whilst on Jan. 21 it was 140 mg. per 100 c.c.

All this time his clinical condition was one of uræmia with mental changes. Urinary output became diminished and sodium sulphate was administered for three days (Jan. 16–18).

The patient then became incontinent and it was impossible to measure urinary output accurately, although the quantity seemed adequate.

Sternal marrow puncture was performed on Feb. 23. It showed normal hæmopoiesis with plasma cells 2·2 per cent.

Uræmic symptoms and high blood-urea remained until he died on Feb. 24.

Serum-proteins on Jan. 16 were albumin 5·1 g., globulin 2·5 g.

The history may be summarized thus: A man aged 72 with slowly increasing pain in hips and legs together with a solitary sacral tumour. General health remained good for a year from the onset of symptoms, at which time uræmia commenced, with death three weeks later.

Bence-Jones proteinuria was present each time a test was made after November, 1946, but serum-globulin was not raised.

FIG. 32.—Anterior view of the tumour at post-mortem examination, seen arising from the sacrum. (× ½.)

FIG. 33.—Lateral view of tumour after vertical section of sacrum. (× ½.)

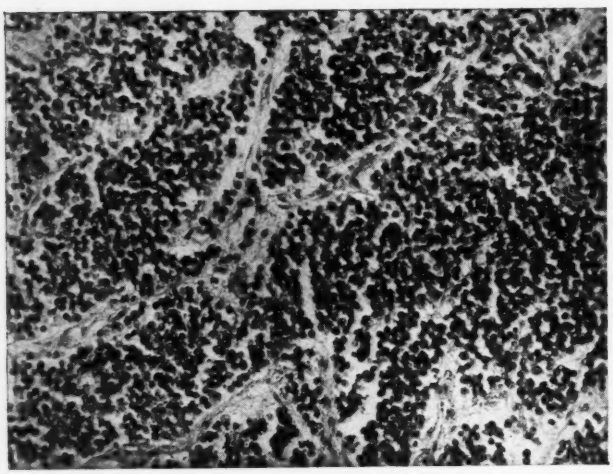

FIG. 34.—Low-power view of tumour histology showing general structure and plasma cells. (× 80.)

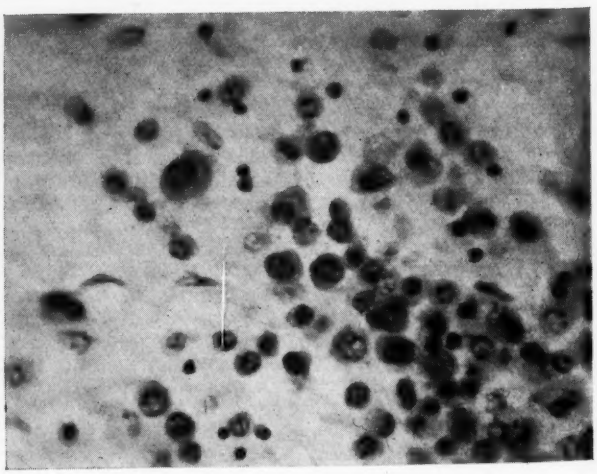

FIG. 35.—Higher-power view of same section as in *Fig.* 34, showing plasma cells, both typical and binucleate forms. (× 335.)

POST-MORTEM EXAMINATION.—This was performed on Feb. 24, 1947, four hours after death, with the following findings:—

The body was that of a well-built old man. There were no external scars or tumours.

Chest.—

Heart: Atheromatous plaques were present on the valves and aorta. Otherwise normal.

Lungs: Some basal congestion.

Brain, Gastro-Intestinal System, Suprarenals, Spleen.—Normal.

Bony Skeleton.—

Sacrum: The entire bone was replaced by a soft fleshy tumour undergoing central necrosis, which extended forwards to become firmly attached to the posterior wall of the rectum. Local infiltration of the rectal musculature had occurred, but the mucosa was not involved. Compression of the lumen with obstruction had occurred. The mass also extended laterally, destroying both sacro-iliac joints and involving the most medial parts of both iliac bones. On the left this extension was only for ½ cm., and on the right the tumour passed posteriorly to the psoas muscle, destroying the bone substance and ending with a well-defined capsule. The mass measured 8 × 8 × 6 cm. (*Figs.* 32, 33).

Microscopically the tumour was composed of a mass of cells of plasma-cell type, ovoid, polygonal, or pyriform in outline, with a nucleus most frequently eccentrically placed and having the typical 'clock-face' appearance. The cytoplasm was plum coloured with eosin, and carmine

with pyronin methyl green. A perinuclear halo, best demonstrated with iron hæmotoxylin stain, was observed in many cells.

The majority of the cells conformed with the above description but giant forms and binucleate forms were also frequently encountered. Cells undergoing mitosis were observed, but were not numerous (*Figs.* 34, 35).

FIG. 36.—External aspect of kidney at post-mortem examination showing hæmorrhagic splashes on pale background. (× ½.)

The power of local invasion of the tumour could be demonstrated in the muscle wall of the rectum.

Bony tissue was almost completely absent and only represented by occasional trabeculæ.

Remainder of Skeleton: The skull was cut in many planes, none of which revealed naked-eye or microscopical tumours.

Thoracic and lumbar vertebræ, ribs, clavicles, right and left humerus, and right and left femur were all examined in section, with negative findings. Bone-marrow scrapings failed to reveal accumulations of plasma cells.

Bladder.—The wall was hypertrophied and the cavity smaller than normal. Chronic cystitis was present. The prostate was enlarged, showing simple hypertrophy microscopically.

Ureters.—Normal. No dilatation was present.

Kidneys.—Weight: right 15 g.; left 14 g. Both were pale grey in colour and externally covered with small red 'splashes'. The capsules stripped easily. The cut surface showed extreme pallor, but the cortico-medullary outline was maintained, although somewhat blurred (*Figs.* 36, 37). The pelvis was not dilated.

Microscopically the kidneys showed little or no change in the glomeruli. No basement membrane thickening or reticulin increase in the tufts could be demonstrated.

Many tubules were blocked with amorphous casts of eosinophilic substance, and when stained by Lepehne's method for hæmoglobin, using benzidine and prohydrol, a negative result was obtained (*Fig.* 38).

Tubular dilatation was not a marked feature, although a number of convoluted tubules were dilated to the size of glomeruli. Casts were found in the distal convoluted tubules, and mainly in the loops of Henle and collecting tubules. The appearance of concentric laminæ was noted and many of the casts were seen surrounded by nuclei, giving the appearance of giant cells. Further reference will be made to this phenomenon later.

The tubular epithelium varied in appearance in different parts of the nephron. The primary convoluted tubules showed granularity of cytoplasm and nuclear degeneration. The post-mortem was performed only four hours after death, and the tissues were fixed immediately. However, it is well known that post-mortem degeneration giving an appearance not unlike that described occurs very rapidly in these cells.

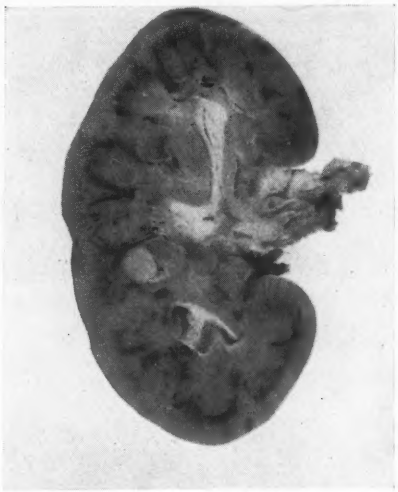

FIG. 37.—Cut surface of kidney. (× ½.)

The loops of Henle and collecting tubules showed compression and thinning in some areas, but were more frequently hyperplastic. Nuclear and cytoplasmic detail was on the whole clear cut in these areas.

The appearance of 'giant cells' around the casts in these tubules was due to the epithelial hyperplasia with

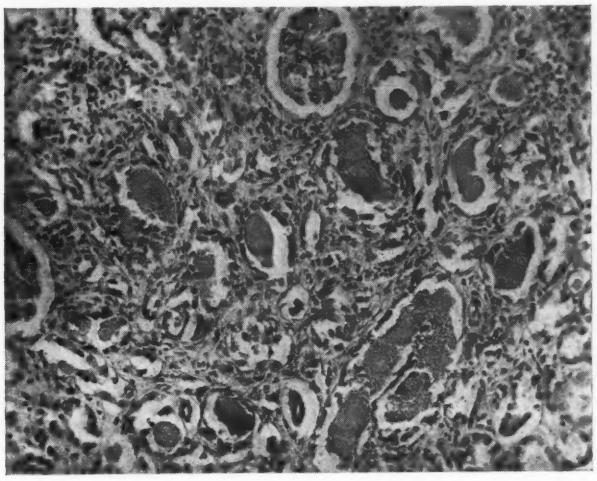

FIG. 38.—Histology of kidney to show casts in the tubules. (× 80.)

subsequent desquamation. Serial sections cut of the kidney showed this fact clearly (*Figs.* 39–41).

Many tubules were collapsed and atrophied. Interstitial fibrous tissue as demonstrated by Mallory's aniline blue was increased in these areas, as was reticulin, although much of the apparent increase of the latter was due to condensation in the collapsed areas. Round-cell infiltration was associated with these changes. There was no evidence of ascending pyelonephritis and sections stained for amyloid were negative.

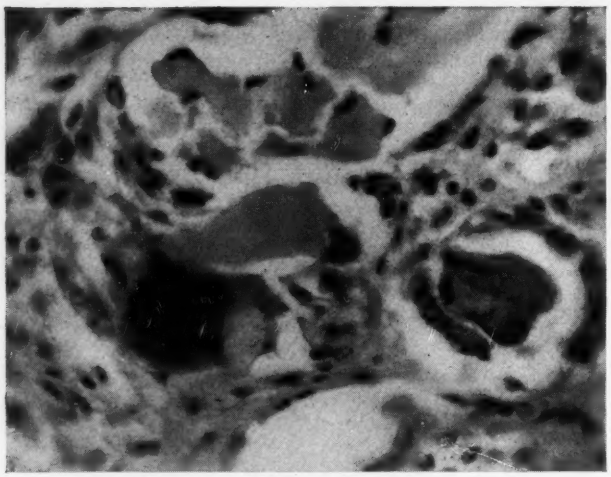

Fig. 39.

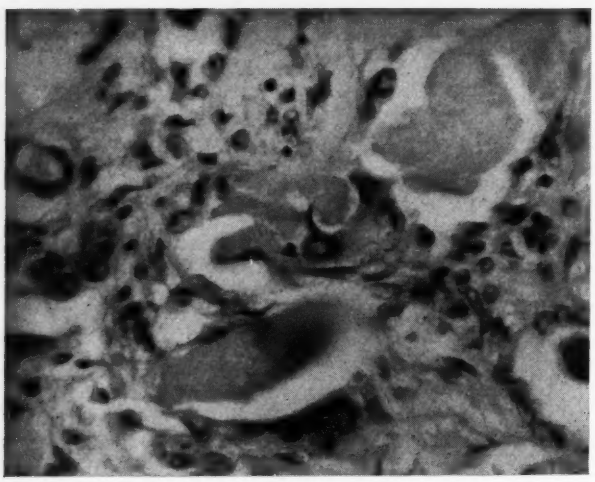

Fig. 41.

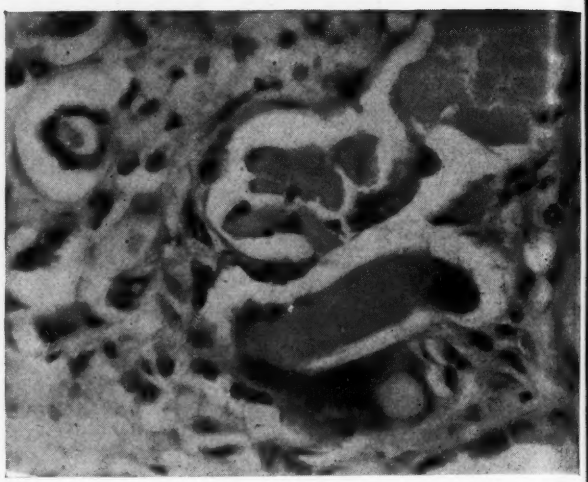

Fig. 40.

FIGS. 39–41.—A series of three sections of kidney cut as serial sections showing hyperplasia of tubule walls simulating giant-cell reaction. (× 335.)

DISCUSSION

The diagnosis of solitary plasmocytoma involving the sacrum is assured. Extensive search for evidence of other tumour masses was made in the cranium, ribs, vertebræ, and less common sites, such as the ilium, femur, and humerus. No tumours were to be seen on section, and smears from the marrow in numerous sites failed to reveal accumulations of plasma cells. It may also be noted that a sternal-marrow biopsy taken just before death showed no abnormality. These facts, taken in conjunction with the negative findings in the radiographs taken of every bone in the body three weeks before death, leave little room for doubt that the sacral tumour was, in fact, a solitary one.

It has been recorded on numerous occasions that Bence-Jones protein is to be found in the urine of many cases of multiple myelomatosis. Geschickter and Copeland (1936) state that it is excreted alone or with albumin in about 65 per cent of cases.

It is generally assumed (Stewart and Taylor, 1932) that Bence-Jones proteinuria is an indication of extensive bone-marrow involvement, and Willis suggests that the finding of this substance in the urine in cases thought to be solitary plasmocytomata, even in the absence of radiographic changes in the other bones, is indicative of multiple myelomatosis. This observation is clearly true in the majority of cases, but it would seem logical to assume that a very large solitary tumour, by the very nature of its extensive bone-marrow involvement, might also lead to Bence-Jones proteinuria.

It should be made clear that the presence of Bence-Jones protein in the urine is not in itself conclusive proof of myelomatosis, as it has been reported in a number of other conditions, notably metastatic carcinoma in bones, multiple sarcoma of bones, senile osteomalacia, fibrocystic disease, chronic lymphatic and myelogenous leukæmia, and polycythæmia rubra vera (Bayrd and Heck, 1947).

These other conditions must obviously be excluded, as was done in this case, before the diagnosis of myelomatosis is made absolute.

The kidney changes in this case closely resemble those found on certain occasions in multiple myelomatosis.

Tubular obstruction of this type also occurs in crush anuria, mismatched transfusions, and sulphonamide poisoning. No further reference need be made to these conditions, however, as at no time did the patient under consideration receive either blood transfusions or sulphonamide drugs.

The kidney changes in multiple myelomatosis have been noted by numerous authors (Löhlein, 1921; Bell, 1933; Forbus and others, 1935; Holman and Hill, 1939; Morison, 1941; and Newns and Edwards, 1944). Bell's is the most comprehensive review, as he describes 11 cases. The description in all the reports is of a normal sized or slightly enlarged kidney, pale in colour, with the cortico-medullary outline blurred but definable. Glomerular changes are minimal. Bell describes thickening of the basement membrane similar to the appearance he found in lipoid nephrosis. Morison describes reticulin increase forming coarse strands in the glomerular tufts.

Tubular blockage is stressed in all cases. Bell noted dilatation of the tubules above the block, but Morison was not able to show this in all his cases. All authors suggest that the casts are of long standing,

as shown by the concentric lamination of many of them. Attention has been drawn to the appearance of 'giant cells' around many casts. Bell believes this is a true macrophage reaction and his opinion is upheld by Holman and Hill. Löhlein, on the other hand, considers that they arise from the tubular epithelium, a view with which Newns and Edwards agree. Morison suggests that epithelial cells lying around the casts may simulate the giant cells. This latter view is borne out in the case under review. The so-called 'giant cells' were always found inside the tubules, and were always associated with epithelial hyperplasia. The photographs of serial sections make this point clear (*Figs.* 39–41).

As to the composition of the casts, most authors are agreed that they do not represent degenerated cells of the tubular epithelium, and the most probable explanation seems to be that of Bell, who suggests a precipitation of Bence-Jones protein or serum-globulin. This view is upheld by Forbus and others.

It will now be seen that most of the renal features attributed to multiple myelomatosis are to be found in the kidneys of the case here presented, and it seems reasonable to suppose that the cause is the same in the two conditions. It is known that Bence-Jones protein had been present in the urine of the patient (H.N.) on numerous occasions, and it seems a most attractive hypothesis to consider this substance as having caused the tubular blockage, with subsequent renal failure and uræmia.

In a search of the literature, no case can be found where exactly similar features are recorded. Numerous cases of so-called solitary plasmocytoma of bone are to be found where Bence-Jones protein was discovered in the urine. None of these cases, however, bear the test of close examination. Either complete radiographs of the skeleton are missing, inadequate post-mortem details are given, or insufficient length of surveillance in order to exclude multiple disease has been allowed.

Two cases only can be found in which renal changes have been noted in probable solitary tumours.

The first is the case reported by Zdansky in 1927, reviewed by Cutler and others in 1936, and allowed by Willis (1941) as a true solitary tumour. In the original paper a single sentence covers the fact that at autopsy amyloidosis of heart and kidney with associated nephrosis were found. No details are mentioned.

The association of amyloidosis with multiple myelomatosis has been frequently noticed. Magnus Levy (1931) reported the phenomenon and Atkinson (1937) collected 40 cases from the literature, whilst Blumberg and Fischbach (1945) reported another one. In these cases, however, tubular blockage was rare and amyloidosis may be disregarded in the case under discussion.

The second case is also reported in the group reviewed by Cutler and others, and is disallowed as being a solitary tumour by Willis in his subsequent review on the grounds of having had Bence-Jones proteinuria on one occasion. In view of the present findings, however, this single factor may not be sufficient to rule out the diagnosis and further review of the case seems to be justified.

The case is one of the Bone Registry of the American College of Surgeons, No. 1148 (Cutler and others, Case No. 16).

The patient was a woman aged 39, who, when seen in January, 1928, had a two-year history of pain in the left hip. When first seen in 1928 she had a swelling over the left anterior superior spine of the ilium and pain radiating down the left leg. In August a biopsy of the left ilium was performed and diagnosed on histological grounds as a plasma-cell tumour (myeloma).

Bence-Jones protein was found in the urine at this time.

Radiographs of the skeleton revealed no other lesion.

In June, 1930, the patient died, and at post-mortem myeloma of the left ilium was confirmed and "gross examination of the skeleton at necropsy revealed no evidence of tumours in other bones". There were signs of pulmonary abscess and chronic nephritis and the authors comment that "unfortunately the clinical analysis from the point of the nephropathy does not indicate whether the kidney changes are referable to the metabolic disturbances associated with Bence-Jones proteinuria (myelomatous nephrotic contracted kidney)". The patient died with symptoms referable to renal failure.

Willis in his subsequent review criticizes the validity of the diagnosis of a solitary tumour in this case, on the grounds of the presence of Bence-Jones proteinuria on one occasion, as stated above, and also on the inadequate post-mortem reference "gross examination of the skeleton at necropsy . . .".

Although insufficient details are available in this case, it is quoted as being the nearest approach which could be found to the case here reported.

The findings in the case recorded may shed some light on the question of the possible relationship between the solitary plasma-cell tumours of bone and multiple myelomatosis.

Whilst in agreement that multiple myelomatosis is of multifocal origin, in the light of the case here reported it seems possible that a solitary tumour of bone represents not a separate and distinct entity, but merely one of these foci. In this case it has produced Bence-Jones protein and even kidney changes. It should perhaps be regarded not as a separate entity, but as a single focus of an essentially similar disease process, usually affecting different bone-marrow sites from the more common multiple disease.

Untoward effects upon the patient possibly depend upon the actual amount of bone-marrow involved, or tumour substance present, whether this is in the form of many small nodules or one large tumour.

SUMMARY

1. A case of solitary plasmocytoma of bone is reported with a brief review of the literature.

2. The principal interest of the case is the association of renal tubular blockage of the type found in multiple myelomatosis with a solitary bone tumour.

3. Bence-Jones protein was found in the urine despite the fact that only one tumour could be demonstrated after careful autopsy.

4. It is suggested that this is not a tumour which has not yet metastasized, nor is it a separate disease

entity, but simply a single focus of a disease usually presenting in multifocal form.

5. It is further suggested that the appearance of Bence-Jones proteinuria may be a result of actual tumour volume, or amount of bone-marrow involved.

My thanks are due to Mr. E. Stanley Lee, the Surgeon in charge of the case; to Sir Stanford Cade, K.B.E., C.B., and Professor R. J. V. Pulvertaft, O.B.E., for much helpful advice; and to Dr. Peter Hansell of the Department of Photography, Westminster Hospital.

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A CASE OF POLYOSTOTIC FIBROUS DYSPLASIA

BY A. C. BINGOLD, LEEDS

'POLYOSTOTIC fibrous dysplasia' is the name first given by Lichtenstein (1938) to a rare skeletal disease characterized by mainly unilateral changes. Subsequently Lichtenstein and Jaffe (1942) defined fibrous dysplasia as "a condition affecting one, several, or many bones, the graver cases of which may present abnormal pigmentation of the skin, premature sexual development, hyperthyroidism, or still other skeletal abnormalities". This wide definition includes both the many variations of fibrous dysplasia, e.g., polyostotic cases, and Albright's syndrome. Albright and his co-workers (1937) described a clinical entity which is distinguished by: (1) bone lesions which tend to be unilateral, and have the histological appearance of osteitis fibrosa; (2) flat brown pigmented areas; (3) endocrine changes, which in girls may cause precocious puberty. Quite recently, however, Albright (1947) has agreed that the best name for the disorder, which he originally had named 'osteitis fibrosa disseminata', is polyostotic fibrous dysplasia.

Clinical Features.—The disease process starts in childhood and ceases spontaneously after puberty. Bony deformities and spontaneous fractures are the outstanding clinical features. The distribution of the changes is predominantly unilateral, but both sides of the skeleton are occasionally affected. Life is not shortened. The oldest patient in Uehlinger's series (1940) died at 67 years of peritonitis. There is no evidence of heredity or the supervention of malignant disease. Endocrine and pigmentary changes may occur. Metabolic disturbances are unusual. Serum-calcium and plasma-phosphorus are normal, but the serum-phosphatase is sometimes raised.

Aetiology and Pathology.—The aetiology is unknown, but it has been suggested that the disease might be due to developmental errors, because it is occasionally associated with other congenital stigmata. Thus Stauffer (1941) reports a case with multiple arteriovenous fistulæ. Boyd (1947) states that "it may be a perversion of the normal process of removal of cartilage by vascular connective tissue preparatory to the formation of new bone". The endocrine disturbances of the syndrome may be explained by postulating a congenital hypothalamic defect (Falconer, 1942). In the cases that went to autopsy the parathyroids were normal.

The essential abnormality is a fibrosis of the marrow, which in the case of the long bones changes the compacta into spongiosa, bends and expands them. The epiphyses remain intact or are only involved late in the disease. Areas of hyaline cartilage frequently occur inside the affected bones.

Histologically the main features are a coarse fibrosis of the medullary canal and atrophy of compacta without any evidence of inflammation or a periosteal reaction. Hæmosiderin may be found inside reticulo-endothelial cells.

Radiological Diagnosis.—The X-ray appearances of the affected bones are diagnostic. They are: (1) Broadening or expansion; (2) Thinning of cortex; (3) Rarefied and apparently trabeculated and cystic appearance; and (4) Secondary deformities.

Complete examinations of the skeleton have revealed that the long bones, especially the femur, are most commonly involved. The base of the skull often shows sclerosis. Uehlinger (1940) states that the bones of the carpus and tarsus invariably escape.

Differential Diagnosis.—The disease must be diagnosed from Paget's disease of bone; and its cousin, Ollier's disease, owing to the occurrence in both diseases of intramedullary cartilage. Hand-Schueller-Christian disease, solitary bone cysts, osteoclastomata, and hyperparathyroidism have also to be considered. The last distinction is important, lest the patient be subjected to an unnecessary exploration of his parathyroids. Hyperparathyroidism affects the whole skeleton, and is characterized by rarefaction and cyst formation. In the glandular type (von Recklinhausen's disease) the serum-calcium is high and the phosphorus low; in the

renal type, where a failing kidney cannot excrete phosphates, the reverse obtains.

Treatment.—Treatment is mainly symptomatic. Fractures are immobilized, deformities can be corrected by osteotomies, solitary lesions may be excised, and weakened bones strengthened by inserting bone-grafts after curetting the site of the lesion. Usually, however, orthopædic appliances, e.g., block leather corsets or walking callipers, are used to give strength and stability to deformed lower limbs. During the active stage of the disease it may be helpful to prescribe aluminium acetate as recommended by Helfet (1940) for the treatment of Paget's disease, but its use is only empirical, as no metabolic disturbance can be demonstrated.

The following is the description of a case of polyostotic fibrous dysplasia seen at the Halifax General Hospital in July, 1947.

CASE REPORT

Mr. J. S. was receiving treatment in the physiotherapy department following a fall on his left hip. On account of the bizarre deformity of his right lower limb he was referred to orthopædic out-patients.

HISTORY.—He was born on July 3, 1877, and was perfectly well until the age of 6 years, when he fell out of bed and injured his right leg. Although he was said not to have sustained a fracture, an inward bending of his right thigh developed. The deformity gradually became worse until he stopped growing at the age of 20 years. He could, however, get about quite well, but was unable to play games. At 20 he started using a stick, and at 30 a stick and a crutch. He was seen by several medical men. One prescribed strong salt baths, another orthopædic boots, a third a leather corset to protect his right knee, a fourth suggested straightening his right leg, a fifth amputation of the whole limb.

He sustained a number of fractures. At 20 years of age he was involved in a tram accident, and broke both forearms. Wooden splints were applied and the fractures united after seven weeks, but he was left with some loss of movement. At 30 he broke his right thigh high up. He was treated on an inclined plane, to which his limb had been secured by bandages and sandbags. After twelve weeks he was discharged walking with crutches. At 39 he sustained another fracture of his hip, but not at the same place. Treatment and after-treatment were similar. The two latter fractures came on gradually and he cannot remember any particular accidents.

His voice broke at 20. At 54 he married a woman of the same age (there are of course no children). Twelve years ago he gradually became deaf in both ears, the left worse than the right, and he has worn a hearing device since.

Family History.—He was one of eight children. Two brothers and one sister are still alive and well. There is no evidence that any member of his family suffered from a bone disease.

ON EXAMINATION.—He is a well-preserved man of 70. His height is 5 ft. 2½ in., his weight 121 lb. He uses a crutch on the right side and a stick on the left (*Fig.* 42). On the right side he wears an orthopædic boot with a raise of 5 in.

Osseous System.—Abnormalities were noted in his right lower limb, his spine, and both forearms.

His right leg shows 5 in. of real shortening, adduction of the thigh, genu valgum, and talipes equinovarus. The circumference of his right thigh, measured 6 in. above the upper border of the patella, exceeds that of the left at the same level by 2½ in. His right calf, measured 6 in. from the upper border of the tibia, is 2 in. thicker than his left. His right femur and tibia feel much bulkier than the corresponding bones on the normal side. His hip is held in 30° adduction without flexion or rotation deformity. It can be abducted to the neutral position, flexed 90°, and rotated normally. There is some telescopic movement of the hip. The great trochanter is raised 1 in. but can be pushed up another 1 in. Flexion of 90° is possible at the knee.

The ankle is held in equinovarus, allowing only 15° of movement. Below this joint the head of the talus is very prominent.

FIG. 42.—Showing the deformity of the patient's right leg, the leather support, his crutch, and stick.

In spite of these extensive deformities his right lower limb is fairly strong, and can even take his whole body-weight. The 8 in. leather cuff around his knee has, however, strangulated a portion of his quadriceps.

His spine shows a marked fixed scoliosis with three curves, compensatory to his short right leg. There is a right dorsilumbar, a left middle and upper dorsal, and a right cervical curve. The associated rotation makes his right loin and his left scapula very prominent.

His left forearm exhibits evidence of an old fracture, with outward bowing of the ulna. Pronation is possible to 90°, supination is full. Flexion and extension of the elbow is from 142° to 45°.

His right elbow has full rotation and flexion and extension from 150° to 30°.

Cardiovascular System.—Pulse 66, regular, brachials hard and thickened, tortuous. Heart not enlarged. Soft systolic apical murmur, not conducted. B.P. 180/105.

Lungs: Normal.

Nervous System.—Normal.

Other Organs.—

Abdomen: Normal except for a left inguinal hernia, controlled by a truss.

Skin: Three discs of tinea versicolor over his sternum. No brown areas.

Genitalia: Normal.

Axillary hair absent, pubic hair scanty. (Several septuagenarians were examined subsequently and it was discovered that this distribution of hair is normal at this age.)

Neck: Normal.

vertebræ, which are sclerotic, all the vertebræ are markedly rarefied and cupped, the changes of senile osteoporosis. The dorsal vertebræ are lipped. A bony bridge spans the gap between the first and second lumbar vertebræ anteriorly. The sacrum is cystic in places.

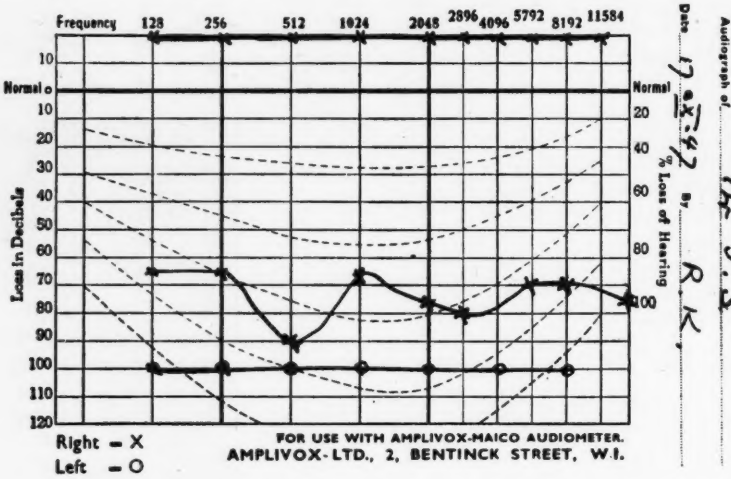
Frequency 128 256 512 1024 2048 2896 4096 5792 8192 11584
Normal
Loss in Decibels
% Loss of Hearing
Audiograph of
Right = X
Left = O
FOR USE WITH AMPLIVOX-MAICO AUDIOMETER.
AMPLIVOX-LTD., 2, BENTINCK STREET, W.1.

FIG. 43.—Audiogram showing complete loss of hearing in left ear.

Eyes: Scleræ grey. Vision good for his age. Fundi show only moderate arteriosclerotic changes.

Ears: Normal drums, prolonged bone conduction, improvement with his hearing aid, and fairly well-modulated voice suggest a diagnosis of otosclerosis. The audiogram (*Fig.* 43) shows that he is completely deaf in his left ear, and has very little hearing in his right. Radiographs of his temporal bones show sclerosis, more marked on the right side than the left, compatible with the above diagnosis.

Urine: Acid. No albumin or sugar. Nothing pathological in deposit.

Biochemical Investigations.—

Aug. 14, 1947: Serum-acid-phosphatase 4·0 K & A Units. Serum-alkaline-phosphatase 6·5 K & A Units. Serum Calcium 11·1 mg. per 100 ml.

Sept. 9, 1947: Plasma-phosphorus 2·7 mg. per 100 ml. Blood-urea 16·0 mg. per 100 ml.

Blood-count (Oct. 17): Hb 85 per cent. R.B.C.'s 4,600,000 per c.mm. C.I. 0·92. Leucocytes 9000 per c.mm. Film shows nothing of note.

X-ray Appearances.—For distribution of the changes *see Fig.* 44.

Lower Limbs: The left lower limb is normal, and in striking contrast with the right, which is characterized by expansion of the shafts of the femur and tibia to about one and a half times the diameter of the left side. The femur has a thin corticalis. Its medulla contains cystic areas alternating with areas of increased density. The neck shows evidence of two old fractures. The shaft has ridden up and has formed a little hollow in the ilium, above what used to be the acetabulum, and is now just a shallow cavity. The bone is markedly adducted. The tibia has a cortex up to 8 mm. wide, and in its medulla, especially in the lower half, are areas of increased density resembling chronic osteomyelitis. Halfway down the shaft there is a round dense shadow about 1 cm. in diameter, presumably a chondroma (*Fig.* 45). The patella is irregular and rarefied. The fibula and the tarsal bones show mainly rarefaction, but the talus and the first metatarsal, its phalanges, and the phalanges of the second and third metatarsals are enlarged and contain cystic and sclerotic areas. The left half of the pelvis is normal; the right side has both cystic and sclerotic areas (*Fig.* 46).

Spine: There is a cervical scoliosis to the right, a high dorsal kyphoscoliosis to the left, and a lumbar scoliosis to the right. With the exception of the 1st and 3rd cervical

Sternum: There are cystic changes from the level of the 4th rib downwards.

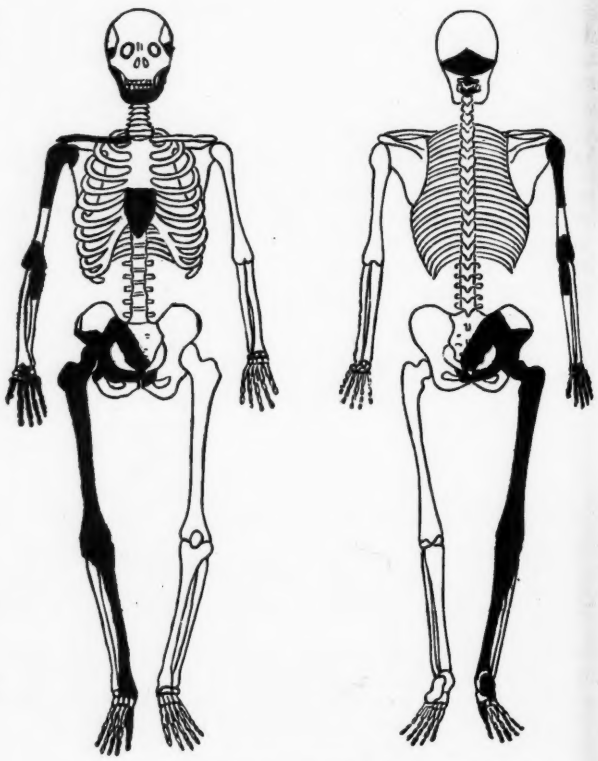

FIG. 44.—Showing mainly unilateral distribution of the skeletal changes.

Skull: The sella turcica is rarefied and expanded, the anterior clinoid processes are rudimentary, the sphenoid is thickened with coarse sclerosis (*Fig.* 47), the occiput shows cystic changes and bony thickening. There are several small cysts on the left side of the mandible.

Left Upper Limb: The left elbow shows osteo-arthritic changes with a loose bony body anteriorly. There is an old, firmly united fracture of the radius at the junction of its upper third with the lower two-thirds. A bony spike projects from the fracture site towards the ulna, one-fourth expands again to resemble the upper portion. The elbow joint is disorganized. There are osteophytes and loose bony bodies surrounding it. The proximal quarter of the ulna resembles the distal quarter of the humerus. The wrist exhibits cysts in the hamate,

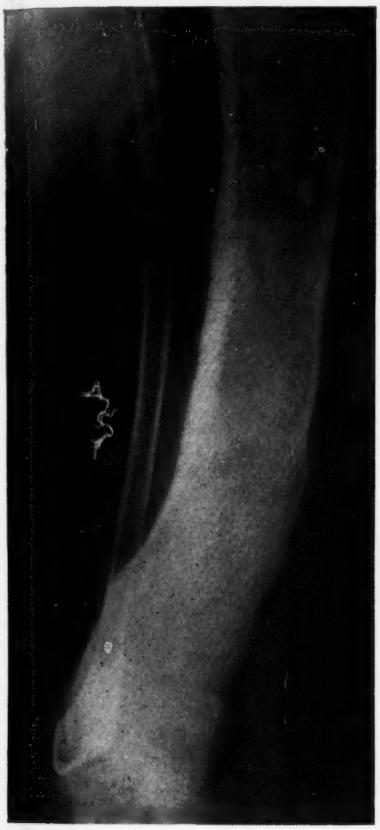

FIG. 45.—Showing lower half of right tibia with thickening of cortex, sclerosis of medulla, and dense round shadow.

FIG. 46.—Pelvis, showing mainly normal left side and sclerosis and cystic changes of right half of pelvis and sacrum. Two old fractures of the femur can be made out. The cortex of the femur is thin and the medulla cystic in places.

which has an old healed fracture at its centre. The heads of the second phalanges of the second to fifth digits contain small cysts.

Right Upper Limb: The clavicle is wider than the left, the glenoid has alternating dense and cystic areas. The humerus is broadened in its upper half, its cortex is thinned, and several cystic areas can be made out. The diameter of the largest cyst is 2 cm. (*Fig.* 48). The succeeding quarter of the bone is normal, but the lower

FIG. 47.—Shallow wide sella turcica with rudimentary anterior clinoid processes. Dense sclerosis of base of skull.

FIG. 48.—Upper end of right humerus, showing cysts and expansion.

scaphoid, and capitate, which has the largest. The metacarpals and the phalanges of the second to fifth digits are broadened and have hardly any cortex. There are cysts up to 1 cm. in diameter in the heads of the metacarpals, the bases and heads of all the phalanges, and the base of the terminal phalanx of the thumb (*Fig.* 49).

FIG. 49.—Showing right hand with cysts in the scaphoid and capitate and affection of the 2nd to 5th metacarpals and all the phalanges. Left hand for comparison.

COMMENT

A typical case of polyostotic fibrous dysplasia has been described. The patient, who is 70, is the oldest recorded in the literature, and demonstrates that the disease does not shorten its victim's expectation of life, but merely causes deformities and spontaneous fractures. Both traumatic and pathological fractures have united in average time in this case. The involvement of the carpus and tarsus, and the sclerosis of the cortex and medulla of the tibia are unusual features. The only endocrine changes were the delayed breaking of his voice at 20 years of age.

The patient has ostosclerosis, and it is tempting to try to establish a connexion between the two afflictions. Radiographs of his temporal bones, moreover, tend to confirm this.

My thanks are due to Mr. Deitch for permission to publish this case; Mr. Hyman for much helpful criticism; Dr. Lewis and Dr. Franklyn for helping me interpret the radiographs; Mr. Lodge and Dr. Kells for the report on the patient's ears; Mr. Mitchell, of the Photographic Department of the Bradford Royal Infirmary, for the reproductions of the X-rays; and Mr. Lumb for the photographs of the patient.

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TANTALUM IN THE REPAIR OF TRAUMATIC SKULL DEFECTS*

BY WALPOLE LEWIN,† M. P. GRAHAM, AND G. B. NORTHCROFT, OXFORD

THE search for suitable materials for repair of skull defects has resulted in the trial and subsequent rejection of many foreign substances. At the outbreak of the Second World War, surgeons were in general agreement that only autogenous bone obtained either from the outer table of the skull or from rib or ilium was reliable. At the same time, the disadvantages of the method were recognized: where rib or ilium is used it entails a second operative field and, for a large defect, a considerable operation to obtain sufficient bone from the donor site; although cosmetic results are usually good, they sometimes fall short of perfection, particularly in the frontal region; also there is a tendency for such grafts to become partly absorbed and for a proportion to become infected. Interest in the possibilities of using foreign materials has been revived over the last four years by the introduction of acrylic resin and tantalum. A preliminary report on the use of acrylic resin at Military Hospital (Head Injuries), Oxford, has previously been made (Small and Graham, 1945); the object of this paper is to describe the results of a trial of tantalum at the same hospital.

Properties of Tantalum.—Tantalum is an element, atomic number 73. Carney (1942) showed that it was well tolerated by the tissues and Pudenz (1943), by studying the fate of such plates in the skulls of cats, demonstrated that tantalum is relatively inert in this situation: it became enclosed in a fibrous tissue membrane without excessive reaction and did not interfere with the growth of the adjacent bony margins. Other workers have confirmed this behaviour of tantalum, with the exception of Delarue, Linell, and McKenzie (1944), who reported serious reactive thickening of the dura mater of dogs if it was inserted subdurally. Tantalum is strong, and a thin sheet ensures adequate protection, yet it is

* Based on a paper read to the Society of British Neurological Surgeons in November, 1947.

† Working with a Grant from the Royal College of Surgeons of England.

sufficiently malleable to be readily fashioned to any required shape in the laboratory and, in so far as minor adjustments are concerned, at the operating table.

These favourable properties suggested a possible application in surgery and Fulcher (1943) was the first to employ tantalum in the repair of skull defects. Woodhall and Spurling (1945) and other surgeons in the United States have reported satisfactory immediate results with this metal.

Tantalum was first used for cranioplasty at Oxford in February, 1944, and was subsequently adopted for the repair of the majority of cases resulting from the campaign in North-west Europe. This paper describes the methods employed and the immediate and late results of the first 130 plates inserted and followed up for a period of at least two years.*

CASE MATERIAL

Between February, 1944, and October, 1945, 130 skull defects in 128 patients were repaired with tantalum, 2 patients who had sustained through-and-through wounds requiring a double repair. Missiles had been responsible in 116 cases: the remaining 12 had received blunt head injuries. The dura was penetrated in 107 instances, intact in 21. *Table I* indicates the approximate sites of the defects.

Table I.—APPROXIMATE SITES OF 130 SKULL DEFECTS

Frontal:		
Involving sinus	33	
Not involving sinus	35	68
Parietal, including vertex		54
Temporal		2
Occipital		6
	TOTAL	130

Indications for Repair.—A visible defect was the main indication, as may be deduced from *Table I*, where it is seen that over half the cases had frontal defects; in addition many of the parietal defects (some of which were really parieto-frontal) extended over the hair line. In some patients a feeling of insecurity or a fear of subsequent damage to the underlying brain was the chief complaint and these were good indications for repair. A few cases had a repair on account of local pain or a beating sensation in the head. We were unable to confirm previous reports that repair of a cranial defect relieves such symptoms as headache and dizziness; repair was never undertaken on account of these symptoms and in the follow-up of the cases treated it was the exception to find a patient who was not complaining to some extent on these scores. There is little profit in a long list of indications for cranioplasty; if the patient requests repair he should not be dissuaded unless there are other overriding considerations, and providing he understands what the operation may reasonable be expected to achieve.

Interval between Wounding and Repair.—This was very variable, the shortest and longest interval in this series being 2 months and 46 months respectively. No repairs were done at the time of the original débridement; although the incidence of early post-operative infection became low, it seemed unjustified in view of the uncertainty of the late results with these plates. Apart from the immediate considerations of the healing of the wound and the condition of the scalp, many of these patients were severely disabled by paralysis and dysphasia and an important stage in their rehabilitation would not be interrupted for a purely cosmetic operation; profound mental change was also a reason for postponing cranioplasty; and the earlier cases had to wait longer since, at the height of battle, beds and time could not be spared for repair operations. However, 48 cases were repaired within four months of wounding. The optimum time for repair will be discussed later.

PREPARATION OF THE PLATE

The method employed is a combination of the metal-beater's craft and some of the processes of dental mechanics, many of the tools and materials in the latter being adapted for this particular purpose. *Fig.* 50 illustrates some of these tools; the boilable

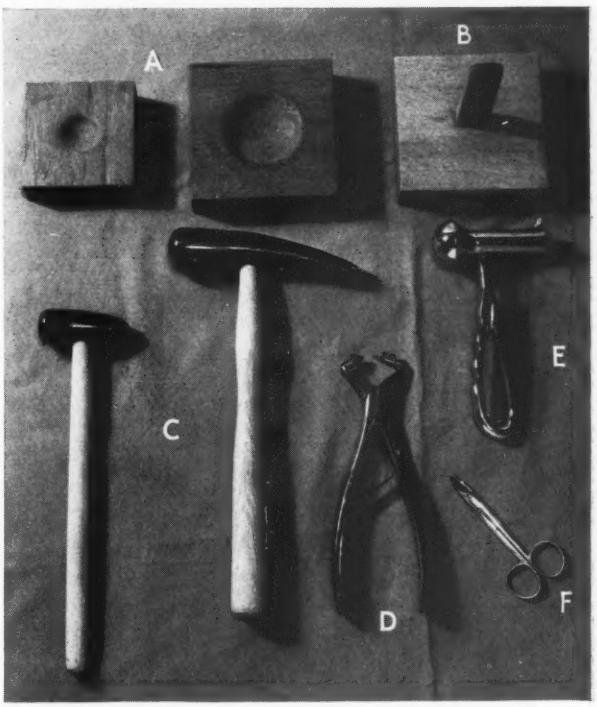

FIG. 50.—Special tools for tantalum plating. A, Boilable teak formers; B, Mounted hardwood peg for shaping orbital plates; C, Horn mallets; D, Dental plate punch; E, Mallet for use in the theatre; F. Dental crown scissors.

teak formers and mallet for use in the theatre were designed specially for this work, the others being obtained through dental firms.

Mechanical difficulties and the time required for the fashioning of the plate to the necessary standard of cosmetic perfection make it an impracticable procedure for the operating theatre and all the cases in this report had their plates preformed to fit accurately on the scalp overlying the defect. However, it was found that whether laid over the pericranium or placed directly on the bone, these plates usually required only slight adjustment at operation.

Obviously the preparation of a plate required to repair a defect involving the supra-orbital region

* In addition to the authors, the following surgeons participated in this work: Sir Hugh Cairns, Lt.-Cols. C. A. Calvert and G. Phillips, Majors W. R. Henderson, O.B.E., and J. Gillingham, M.B.E.

involves a more complicated procedure than that required for a simple defect of the vault, since the plate must cover not only part of the forehead but also parts of the temple and orbital roof. Again, although the average size of defect in this series was about 6 cm. in diameter, some very large plates were required (the largest measured 14 cm. × 12 cm.) and here a variation of technique was necessary. The preparation of plates for repair of small defects of the aid of this pattern, the plate is cut from 0·015-in. sheet tantalum with plate shears. The beating of the plate into the required shape takes place with the patient in the laboratory, so that the plate can be frequently tried on the patient's head until the contour conforms correctly with the surroundings and until there is uniform pressure on the skin all round the periphery of the plate when it is held firmly on the scalp. The materials used for this stage are horn

Fig. 51.

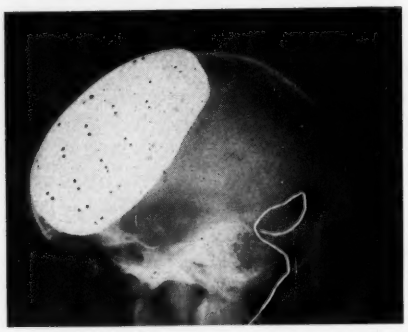

Fig. 53.

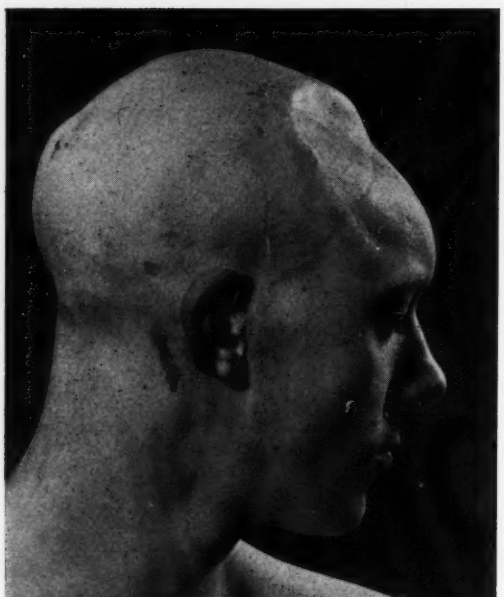

Fig. 52.

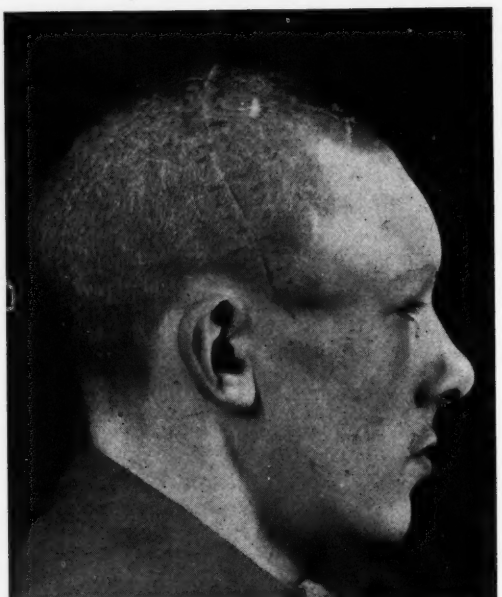

Fig. 54.

FIGS. 51–54.—*Case* HN./8984. Lateral radiographs and photographs before and after plating to show result obtained in a large defect of simple contour by the method described in the text.

simple contour, of large defects of simple contour, and of defects of complex contour will therefore be described under separate headings.

Small Defects of Simple Contour.—This includes any defect of less than 8 cm. in maximum diameter not involving the orbital margins. After shaving the head, the outline of the bony defect is defined on the skin with indelible pencil and checked by careful palpation. A sheet of thin paper is held down firmly on the scalp and a ' rubbing ' made of the pencilled outline. The paper pattern is then cut out allowing an extra $\frac{1}{4}$ in. margin all round. With mallets and a set of teak blocks which have saucer-shaped central depressions of varying diameter and depth.

Large Defects of Simple Contour.—Large plates may be formed by the method described above, but considerable experience is required in the art of metal beating. Better results may normally be obtained by the following method :—

The margins of the defect and the centre line of the head are first marked in indelible pencil. An impression in plaster-of-Paris is then taken, including not only the area of defect but the corresponding

area of the opposite side. After applying a separating medium—oil, talc, etc.—to this impression, a cast is poured in the same material; the pencil markings will be transferred to this model. Dental wax is built up in the depressed area to reproduce the lost contour and continued until, when viewed from all angles, the shape conforms with that of the contralateral area. A reverse of this waxed-up area is then made by taking an impression in 'stone-hard plaster', which should extend about an inch beyond the wax margin.

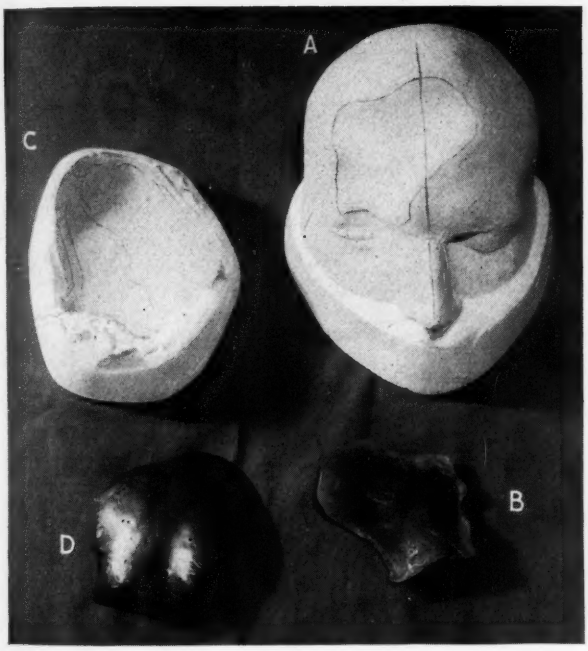

FIG. 55.—Stages in the preparation of a plate for a defect of complex contour. A, Plaster model; B, Wax 'build-up'; C, Counter die; D, The finished plate.

Using a paper pattern, the tantalum plate is cut and beaten roughly to shape in the same way as described for small defects, the waxed area on the model being used as an additional guide. Further shaping is carried out by beating into the concave stone counter. Final adjustments are made after trial fitting on the scalp. *Figs.* 51–54 illustrate the cosmetic result obtained with a plate made by this method.

Defects in Areas of Complex Contour.—These are the defects of the frontal region which involve one or both orbital margins. There were 25 such cases in this series.

The laxity and the variation in thickness of the tissues of the supra-orbital and frontomalar regions, compared with the uniform scalp cover of the upper frontal region, are big disadvantages in the shaping of preformed plates, and considerable judgement and experience are required to achieve a satisfactory cosmetic result; even so, the ultimate result is always somewhat in doubt.

We have found it best to proceed as for large defects, using a waxed-up plaster cast and counter die; the curved and rounded edge of the plate which will replace the lost orbital margin is shaped by beating over hardwood pegs mounted on blocks (*Fig.* 50). Especial care is required in this type of plate to obtain a very accurate fit on the scalp before operation; a snug fit is particularly important where the plate is going to take purchase on the frontomalar buttress. *Figs.* 55, 56 illustrate the steps in fashioning a plate by this method, and *Figs.* 57, 58 the cosmetic result obtained in that particular case, which was not, as will be appreciated, by any means perfect. *Figs.* 59–61, however, show the very good result that can be obtained even in a bifrontal complex plate.

It is important in the final preparation of all plates to clean them thoroughly with nitric acid, followed by several washings. Sterilization is by

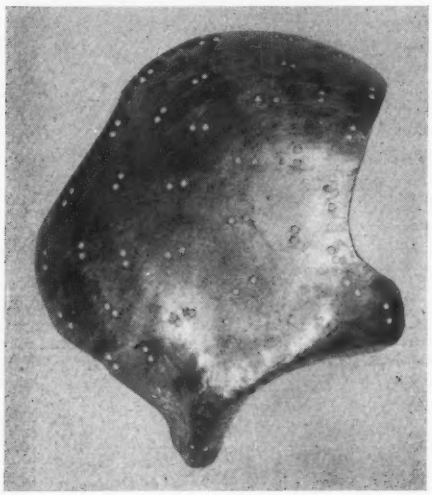

FIG. 56.—Details of the plate. Note the restored orbital margin and the paired holes in the plate.

boiling. It will be noticed that all plates have a peripheral row of paired perforations and variously placed central holes (made with a dental plate punch); these allow stitching to the pericranium or wiring to the bone and also escape of C.S.F. through to the subgaleal space, where it can be absorbed. In addition, the anterior and posterior margins of the plate are indicated to assist the surgeon in fitting the plate.

OPERATIVE TECHNIQUE

Pre-operative.—Chemotherapy was employed in all cases and in the absence of complications continued until the third day post-operatively. Sulphadiazine, 2 g. 4-hourly, was begun 24 hours before operation and intramuscular penicillin, 15,000 units 3-hourly, begun on the morning of operation.

Phenobarbital was also given as routine: for patients who had never had epilepsy, 1 gr. twice daily by mouth, begun the day before operation; declared epileptics already on anticonvulsants would have the dose increased over the operation period.

Operative.—The condition of the scalp overlying the defect was of prime importance. A scar which appeared satisfactory might not remain so under the increased tension resulting from insertion of the plate, and the aim was to obtain total cover of the plate with full-thickness scalp which had a good blood-supply. To this end, a preliminary excision

of a wide scar was required in some cases, the tantalum insertion being delayed for a further six weeks; also, wherever possible, a flap was turned about the defect so as to avoid the possibility of delayed healing in an incision directly over the plate. where the dura was nicked, with escape of cerebrospinal fluid. We purposely left as much tissue as possible on the under-surface of the scalp, and where it seemed thin sometimes applied plaques of fibrin foam to its under-surface. Obvious dural rents were

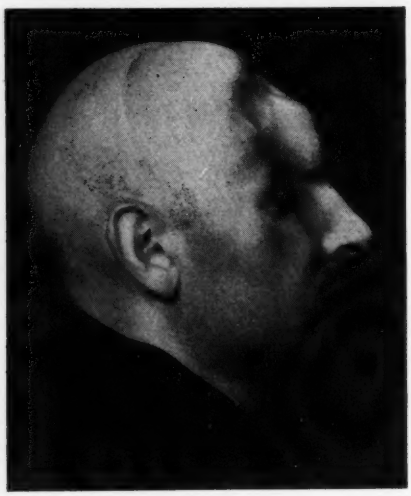

Fig. 57.

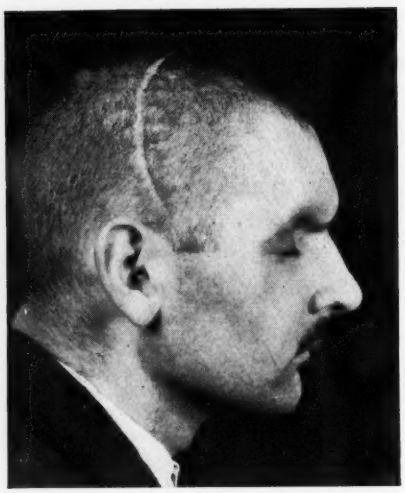

Fig. 58.

FIGS. 57, 58.—*Case* HN/10958. To show the cosmetic result obtained with the plate illustrated in *Fig.* 56. Despite the apparent perfect fit before operation, it is seen that the orbital margin is too high. This plate was later removed and another inserted, this time with a satisfactory result.

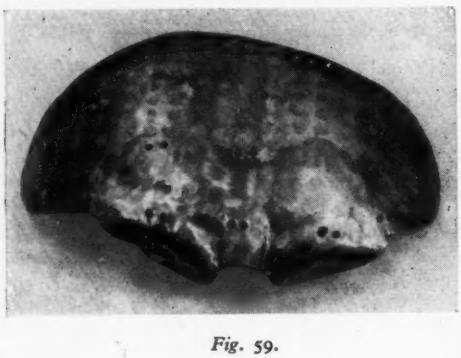

Fig. 59.

FIGS. 59–61.—A good cosmetic result obtained with a bifrontal complex plate. (In this particular case the tantalum employed consisted of thin sheets which were spot-welded as sheets of the correct thickness were not available at the time.)

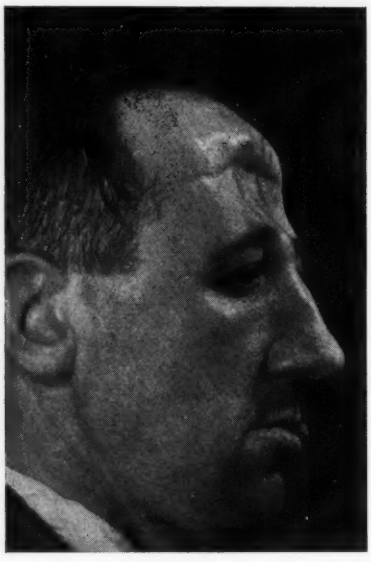

Fig. 60.

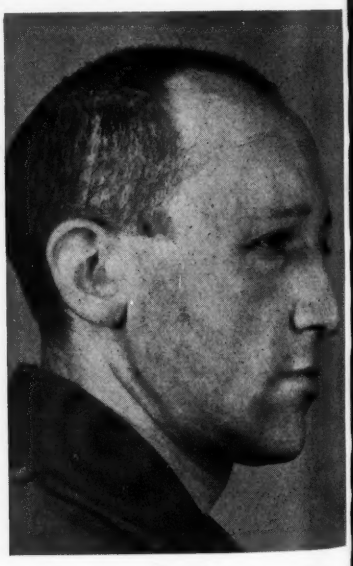

Fig. 61.

When rotation flaps had been used to close the scalp at the time of the original débridement, considerable ingenuity had to be exercised when repairing the defect to fashion a flap with due regard to the main blood-supply of the scalp. The incision could only be planned when the whole head had been shaved, a procedure which frequently disclosed scars of unsuspected length, but also showed where the main arteries of supply of the scalp were situated; whenever possible these were left intact.

In reflecting the scalp from the dura or false dura over the defect there were, inevitably, some cases sutured or repaired with a pericranial or fascia lata graft; the frequency of post-operative collections of C.S.F. beneath the galea suggests that the most careful repair is required in all cases where there has been any nicking of the dura.

The method of insertion of the tantalum plate was simple. It was essential that the plate fitted easily without forcing, and that its margins met the surface of the pericranium or bone accurately everywhere. Minor adjustments were sometimes necessary at the table. If an alteration in size was required or a small V had to be cut to make the plate lie

better, dental crown scissors were used; their small blades cut the plate without distorting it. Final fitting of the margins of the plate could be done by tapping gently with the special mallet shown in *Fig.* 50.

Fixation of the plate varied with the surgeon and the local conditions: it was either sutured directly on to the pericranium (*Fig.* 62), or placed directly

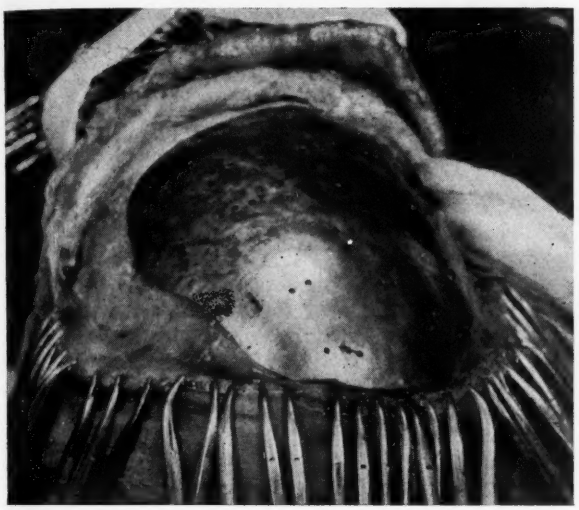

FIG. 62.—Onlay fixation of a tantalum plate by silk sutures to the pericranium. The plate fits snugly on the pericranium and overlaps the bony defect.

on the bone under pericranial flaps which, after suture to the plate, would partly cover it. In some cases a fascia lata graft was stretched over the plate and stitched to the pericranium. Five cases had a step cut in the bone to form a bed in which the plate could lie. Silk was the suture material employed except for the plates in the low frontal region, where fixation was sometimes difficult, when it was reinforced by one or two steel wire sutures between the plate and the bone.

In retrospect, we realize that whereas this simple method of insertion and suture is quite satisfactory over the vertex, it is not so in the low frontal and lateral orbital regions: here the plates have a tendency to slip and a far more adequate direct fixation to bone is necessary. Tantalum is heavy (sixteen times heavier than acrylic resin) and migration of the plates in this situation is partly due to gravity. In all, 15 plates have been removed at this hospital up to July, 1947, including cases not in this series since their repairs were done less than two years ago, and 4 of these have been orbital plates which had slipped downwards across the orbit. We consider now that if tantalum is to be used in this situation it should be applied directly to the bone, its margin should be fitted accurately and held in place securely by wiring to the bone all round. It must be emphasized that wire sutures are only employed to secure an already well-fitting plate and any attempt to use them to hold down an ill-fitting margin will lead to failure. In some cases additional security may be required by counter-sinking in the bone.

Again, at the time when these operations were performed, tantalum wire was not readily available, but it will be used exclusively in fixing tantalum plates in the future, for, as Venable (1942) has pointed out, a combination of two metals should never be used, for even if inert separately they may act together as a destructive battery.

The scalp was closed in two layers of interrupted silk sutures without drainage, after the wound had been dusted with penicillin-sulphamezathine powder, 5000 units/gramme.

Associated Brain Operation.—An intracerebral operation at the time of repair was performed in 28 cases. Sixteen had metal fragments, bone chips, or scar tissue removed, and a cerebral abscess was excised in 3 cases. Fascial repair of the dura was carried out in 22 cases, some of them for tears in the dura over the paranasal sinuses.

Post-operative.—Beyond the moderate pressure of a crêpe bandage over the usual dressings, no special precautions were taken to prevent fluid collecting beneath the scalp, and with few exceptions, despite careful hæmostasis of the wound at the time of closure, serosanguineous fluid collected beneath the scalp flap and required aspiration on the day following operation. These collections were more frequent than after other cranial operations and in some cases obviously consisted of cerebrospinal fluid; over large plates as much as 100 c.c. would be aspirated, but the average amount was about 10 c.c. However, only a few cases required more than one aspiration and in only 3 were they of any moment. Two of these had persistent collections of cerebrospinal fluid beneath the scalp which required repeated aspiration for six weeks before they settled down, and the third case persisted for fifteen months until re-operation. This complication will be referred to later.

All the stitches were removed by the fifth day and the patients were up within a week.

IMMEDIATE RESULTS

These were very satisfactory; first-intention healing of the wound took place in all cases but one, and with three exceptions the cosmetic result was good. Of the 128 patients, 110 had a completely uninterrupted convalescence. There were no deaths.

The early complications, whether directly related to the plate or not, are listed in *Table II.* Three

Table II.—EARLY POST-OPERATIVE SEQUELÆ

Persistent collection of C.S.F. beneath scalp		3
Pressure necrosis of scalp and subsequent infection		1
Meningitis		1
Unsatisfactory cosmetic result		3
Epilepsy within 14 days after operation:		
With epilepsy before operation	5	
First attack after operation	5	10
TOTAL		18

cases, already referred to, had persistent collections of subgaleal C.S.F. The one case to become infected was a complex plate inserted in relationship to an eye socket; the conjunctiva was nicked during the dissection of the scar tissue above the eye socket, and despite suturing, a C.S.F. leak developed post-operatively; infection and pressure necrosis of the adjacent scalp followed and the plate had to be removed. A full case report is given later.

In another case a coliform meningitis developed post-operatively. This was a complication of the

cerebral part of the operation and not of insertion of the plate; the patient had a frontal sinus wound with an aerocele communicating with the ventricle, and at the cranioplasty deep bone chips were removed from the frontal lobes and the torn dura mater behind the frontal sinus repaired with fascia lata. The infection settled down and the plate remained satisfactory.

In 3 cases, all frontal defects, the cosmetic results was clearly unsatisfactory, and 2 of these shortly afterwards had fresh plates inserted with a satisfactory result.

The significance of post-operative epilepsy in these cases will be discussed later on; there were no cases of post-operative status epilepticus.

LATE RESULTS

The follow-up of these cases was conducted primarily by questionnaire. The patients were asked if the plate was satisfactory as regards appearance and comfort and to describe any complications that had arisen. In addition they were asked to complete a special Head Injury Form devised by Dr. Ritchie Russell for the follow-up of such patients. Supplementary information was later obtained where necessary, from the patient's doctor and from any hospitals he may have attended. Some patients were seen personally and several were visited in their homes and were helped to complete the questionnaire by members of the British Red Cross Organization, who often added very helpful observations. Except for 4 members of the Allied Forces who had returned to their own countries and could not be traced, replies were received from all. *Table III* summarizes the results of the follow-up scheme.

Table III.—FOLLOW-UP DETAILS (128 CASES)

		Cases
Follow-up (time after insertion of plate):		
Over 3 years	4	
2½–3 years	38	
2–2½ years	79	121
Deaths (status epilepticus 2; pneumonia 1)		3
Untraced (non-British personnel)		4
TOTAL		128

Three patients died within the period of follow-up. One developed status epilepticus and died shortly after an operation at which the tantalum plate was removed; this case is therefore included in the discussion on late complications. The deaths of the other two patients were unconnected with the plate. One remained well for a year after the operation and then succumbed to pneumonia; the other died in a mental hospital in status epilepticus one year after plating. He had had an extensive penetrating frontal sinus wound complicated by a streptococcal cerebral abscess; at the time when the tantalum plate was put in, the abscess was excised and the dura repaired with fascia lata. Five months later he had an attack of meningitis and seven months after that suddenly went into status epilepticus, from which he died. An autopsy performed some days after death was of limited value as the brain was decomposed, but the tantalum plate was reported as being quite satisfactory. (Headache and high temperature shortly before the onset of status epilepticus suggest that there may have been a recurrence of meningitis.)

Of the remaining 125 patients, 94 have reported that their plates are satisfactory as regards both appearance and comfort; 4 patients are untraced. *Table IV* classifies the complications reported by the remaining cases, including the first of the 3 deaths referred to above.

Table IV.—LATE SEQUELÆ

a. Infection:			
With removal of plate		5	
With retention of plate		4	9
b. Intracerebral complications requiring removal of plate:			
For cerebral abscess (plate satisfactory)		1	
For localized hydrocephalus		3*	4
c. Cosmetic result not entirely satisfactory:			
Frontal		5	
Temporal		1	6
d. Local pain or irritation:			
Frontal		4	
Parietal		4	
Temporal		1	9
	TOTAL		28

* 1 death.

Plates Removed.—Eleven of these 130 plates have been removed to date—2 were cosmetic failures; 5 were removed on account of infection; and 4 for an intracerebral complication.

The main points for discussion in assessing the results of tantalum repair are: (1) the general comfort and cosmetic results; (2) the relationship to infection and, arising from that, the results of using tantalum in frontal sinus wounds; (3) intracerebral complications; (4) the relationship to epilepsy.

APPEARANCE AND COMFORT

It is not easy to assess cosmetic results on the results of a questionnaire alone since, with these wounds, there are sometimes reasons other than the bone defect to account for the poor appearance which will not always be correctly interpreted by the patient; on the other hand a good report from the patient himself means that the main object of the operation has been achieved.

In 3 cases the result was recognized as a failure in the early post-operative period; all were complex frontal sinus plates and 2 of them were replaced, one two months and the other three months later, this time with a satisfactory result; tantalum was used again in one case and acrylic resin in the other. Six other patients have reported slight disfigurement in the absence of other complications and this presumably became apparent at a later date when the scalp finally settled down. The complaint was usually of a slightly sunken appearance, or of an edge which was prominent; all save one were frontal cases and in that case, a temporal defect, it is probable that the missing temporal muscle was as much to do with the poor result as was the shape of the plate.

In general therefore, the one-stage preformed-plate method has produced very good cosmetic results, though its limitations are seen in the frontal region where a perfect result is demanded; 8 of the 68 frontal plates (12 per cent) in this series were not entirely satisfactory, but only 3 of these were definite failures.

It has been mentioned earlier that the cosmetic results of some of these difficult complex plates obtained by the preformed one-stage method described above cannot be guaranteed. There is little doubt that a two-stage method in which a direct impression of the bone defect is taken at the first operation would improve the results; but then tantalum would have no advantage over the more

readily adaptable acrylic resin which gives a perfect cosmetic result. In addition there is some risk of low frontal tantalum plates slipping unless special precautions are taken. These two factors suggest, therefore, that tantalum is not always the method of choice for this region.

The great majority of the plates have been comfortable, but 9 patients (7 per cent) have reported, in the absence of other complaints, local pain or discomfort which is directly referable to the plate. The reports usually referred to "tenderness along the plate edges at times", "periodic pain", or "discomfort"; none of these symptoms appears to have been severe.

INFECTION

In the first place it is important to know the late results of tantalum plating in cases where there has been previous wound infection, meningitis, or cerebral abscess and to determine how long one should wait after such a complication before undertaking the repair.

Wound Infection before Plating.—Delayed healing due to infection of the original wound was present in 31 cases of this series. As shown in *Table V*, several of these patients came to primary operation late or had no initial débridement at all. Wounds involving the temporal muscle or cases with multiple peppering of the scalp were especially liable to become infected and in some cases extradural pus formed. Complete breakdown with fungus formation occurred in only 5 instances.

It will be seen from *Table V* that the late results after repair of these cases were very satisfactory; 4 cases were untraced, but of the remaining 27, only 1 became infected after plating. It is interesting to observe that this latter case was plated just two months after healing of a cerebral fungus; the case history (*Case* 7) is summarized below.

Meningitis before Plating.—*Table VI* summarizes the details of 6 cases which were complicated by meningitis after wounding. The criteria of meningitis were clinical signs and symptoms and over 600 white cells in the lumbar C.S.F. The earliest case to be repaired was a patient with a frontal sinus wound who had had delayed wound healing due to recurrent stitch abscesses, and three attacks of meningitis; he had a dural repair and

Table V.—WOUND INFECTION BEFORE PLATING (31 CASES)

TIME OF INITIAL OPERATION AFTER WOUNDING	CASES	HEALING TIME		
		<6 Weeks	< 3 months	> 3 months
< 72 hours	19	7	11	1
> 72 hours	7	1	6	0
No operation	5	0	3	2
TOTAL	31	8	20	3

RESULTS OF PLATING

INTERVAL BETWEEN HEALING AND TANTALUM REPAIR IN MONTHS	CASES	FOLLOW-UP		
		Satisfactory	Unsatisfactory	Untraced
3	8	6	1*	1
3–6	9	9	0	0
6–12	8	7	1†	0
12	6	4	0	2
TOTAL	31	26	2	3

* Removal of plate 27 months later for infection.
† Hydrocephalus (no infection).

insertion of a tantalum plate one month after final healing of the wound and only eighteen days after subsidence of his third attack of meningitis. At follow-up all these patients were satisfactory.

Cerebral Abscess before Plating.—Only 7 cases of cerebral abscess were encountered in the series (*Table VII*). In every case a dural repair was carried out before inserting the plate, and in all except one either the intracerebral abscess or its

Table VI.—RESULTS OF PLATING AFTER MENINGITIS (6 CASES)

CASE NO.	WOUND					MENINGITIS		TANTALUM REPAIR		RESULT OF FOLLOW-UP	EPILEPSY
	Site	Débridement within 72 hr.	Dura Pen.	Healing	Retained F.B.	Duration in Days	Organism	Interval after Meningitis	Other Operative Details		
9464	Frontal sinus	+	+	1st intention	Bone	8	G+ diplococci and G− bacilli in films. Sterile.	4 mth.	Removal of F.B.s. Fascial graft to dura	Satisfactory	+
1030	Frontal	+	+	1st intention	—	17	Sterile	10 weeks	—	Satisfactory	—
9359	Frontal sinus	+	+	Delayed	Bone	3 attacks in 3 mth.	Sterile	18 days	Fascial graft to dura	Satisfactory	—
2958	Frontal sinus (ventricle)	+	+	Delayed	Bone	11	Coliforms	3 mth.	—	Satisfactory	—
9044	Occipital	+	+	Delayed	—	14	*S. albus* ? *Str. hæmolyticus*	12 mth.	—	Satisfactory	—
9963	Occipital	+	—	1st intention	—	15	*S. aureus*	11 mth.	Fibrin film to dura	Satisfactory	—

F.B. = Foreign Body.

resultant scar was excised at the time of plating. The high incidence of epilepsy and the close relationship of cerebral abscess to late débridement of the primary wound and delayed healing should be noted. The follow-up was satisfactory in 6 of the cases and the death of the seventh case from status epilepticus could not be attributed to the plate.

Table VII.—Results of Plating in 7 Cases of Cerebral Abscess

Case No.	Wound				Abscess Treatment prior to Plating	Organism	Tantalum Repair		Result of Follow-up	Epilepsy
	Site	Time of Débridement	Healing	Retained F.B.			Interval after Healing	Other Operative Details		
7428	Fronto-parietal	No opn.	Delayed	Bone and Metal	Discharged through wound	?	6 mth.	Cerebral scar containing microscopical abscesses and F.B.s. excised. Fascial graft to dura	Satisfactory	+
9132	Frontal	3 days	Delayed	—	No details	?	3 mth.	Fascial graft to dura	Satisfactory	—
10120	Temporal	No opn.	Delayed	—	Extra- and intra-dural abscesses drained. Later, excision lateral wall of abscess	?	9 mth.	Excision of abscess and dural graft	Satisfactory	+
8043	Vertex	24 hr. (incomplete)	Delayed	Bone	Drained	Coliforms, *S. aureus*, *S. viridans*	12 mth.	Excision of cerebral scar and F.B.s. Dural graft	Satisfactory	+
2367	Vertex	3 days	Delayed	Bone	Abscess drained and F.B.s removed 3 years after wound	Staphys. Non-hæm-strep.	10 mth.	Excision of abscess scar and F.B.s. Fibrin film to dura	Satisfactory	+
8664	Frontal	3 days	Delayed	Bone	Aspiration followed by excision and marsupialization of 2nd abscess	*S. aureus*	6 mth.	Excision of abscess scar and F.B.s. Dural graft	Satisfactory	+
10472	Frontal sinus	24 hr.	1st intention	Bone	Aspiration 4 mth. after wounding	*Str. hæmolyticus*	—	Excision of abscess and dural graft 4 mth. after initial aspiration	Died*	+

* Died in status epilepticus 1 year later; ? meningitis; tantalum plate satisfactory. F.B. = Foreign Body.

These results suggest that with modern chemotherapy a history of previous infection does not preclude a successful tantalum repair. Although on general surgical principles one would defer the operation under such conditions for at least six months, it appears that if an early repair is desirable it can be safely carried out three months after final healing of the wound.

Infection after Tantalum Repair

Six tantalum plates were complicated by frank infection with formation of extradural pus, and 4 of these had to be removed; chronic infection around the plate in 1 case was disclosed at operation for traumatic epilepsy and the plate was accordingly removed. A further 2 cases have had symptoms strongly suggestive of low-grade infection which has subsided spontaneously without treatment; they are, nevertheless, included in this group. The histories of these 9 cases are summarized below:—

Case 1.—*HN*/9265, July 20, 1944. A through-and-through left frontotemporal wound involving the frontal sinus and left orbit which necessitated evisceration of the left eye and partial excision and suture of the lid margins. The mucous membrane of the left frontal sinus was removed. The dura was left open and the wound healed by first intention. Post-operatively the patient had a temporary C.S.F. rhinorrhœa.

Seven months after wounding cranioplasty was performed. During the dissection above the eye socket, the conjunctival sac, which had become drawn up by scar tissue, was opened; this communication was closed with a stitch. No dural tear was found. A complex tantalum plate was fitted to fill the defect which involved the lateral wall of orbit and the supra-orbital margin, and was anchored by pericranial stitches and steel wire sutures through the bone.

The patient developed a C.S.F. leak through the eye socket post-operatively which macerated the tissues over the rather sharp edge of the tantalum forming the new external angular process. Pressure necrosis and subsequent infection with *Ps. pyocyanea* followed and the tantalum became visible in the eye socket. An attempt made to conserve the plate failed; it became displaced down across the orbit and finally, three months after insertion, it had to be removed.

Case 2.—*HN*/9863, Aug. 10, 1944. A through-and-through left frontal–right temporal wound with fractures extending across both frontal sinuses. First-intention healing followed the initial wound treatment.

Three months later, through a coronal scalp flap, the residual bone-chips were removed from the left frontal lobe; no tear was found in the dura in relationship to the sinuses. Two tantalum plates were inserted, one to the left frontal region and one to the right temporal region, and fixed by stitching to the dura and stretching over each a sheet of fascia lata which was sutured to the pericranial margins. The patient made an uninterrupted convalescence.

One month after leaving hospital and seven weeks after the cranioplasty, he developed a severe head cold which was soon followed by a swelling of the forehead over the

plate and œdema of the left eye and cheek. He was readmitted to another hospital where incision over the swelling disclosed pus. Ten days later, the frontal tantalum plate, which was bathed in extradural pus, was removed at Killearn Neurosurgical Unit by Mr. Paterson. The wound healed promptly.

A year later a fresh tantalum plate was inserted at the same hospital. The plate in the right temporal region has remained satisfactory.

This is a very interesting case where it seems clear that the relationship of the plate to a damaged frontal sinus determined infection, a plate elsewhere in the skull remaining satisfactory.

Case 3.—*HN*/9418, Aug. 7, 1944. A penetrating right parietal wound in which at the initial operation the dura was closed with silk sutures. The wound healed by first intention. A tantalum plate was fitted to the defect ten months later.

The patient made an uninterrupted convalescence, but six months after this operation, whilst recovering from a heavy cold which was attended by pyrexia and malaise, he struck his head sharply on the doorway of a taxi over the site of the plate. He did not lose consciousness and there was no scalp injury. However, one week later, a fluctuant swelling appeared over the plate and slowly increased in size. Six weeks after the injury he was admitted to Bangour Neurosurgical Unit and the area of plating explored by Mr. G. L. Alexander. The plate had slipped and was surrounded by pus which on culture grew *Staph. aureus*. It appeared that the tantalum plate was also a little bent, presumably from the blow. The patient made a rapid recovery after removal of the plate and ten months later it was re-inserted.

Case 4.—*HN*/11378, April 7, 1945. A right vertex bomb wound in which the sagittal sinus was torn but elsewhere the dura was intact. At the primary operation

FIG. 63.—*Case* 4. Infected tantalum plate. The scalp is œdematous and a sinus at the posterior margin of the old incision is filled with granulations and is discharging pus.

the dura was opened and necrotic brain tissue was sucked out. The dural incision was closed and the wound healed by first intention. Tantalum repair was carried out six months later, and the plate anchored extrapericranially.

The patient reported eleven months later that he felt that the plate had slipped, and radiographs taken then showed that the fit was certainly not accurate but unfortunately there were no immediate post-operative films for comparison. He returned home, but four months after this reported that a fluctuant swelling had appeared over the plate; shortly afterwards pus discharged through a small sinus. This was treated conservatively with penicillin over the next two months and the infection subsided.

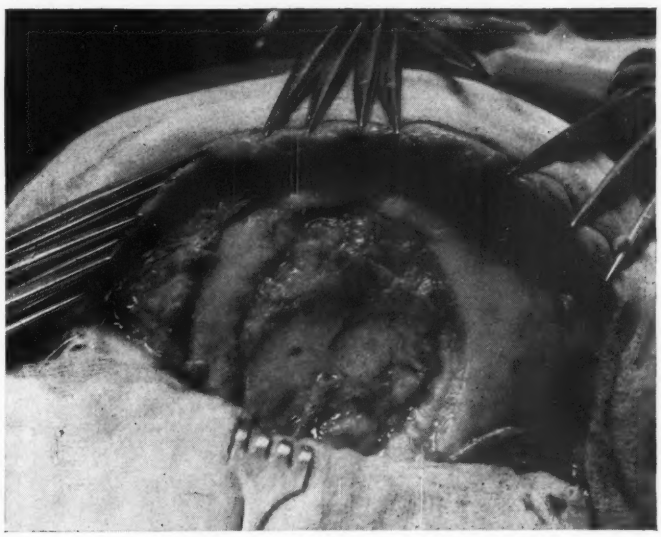

FIG. 64.—*Case* 4. Operative findings. The tissues are œdematous, the bony defect is covered by unhealthy granulation tissue and pus, and the bony margins are infected.

It recurred three months later and this time considerable quantities of pus discharged through a sinus at the posterior margin of the wound, which was filled with pouting granulations (*Fig.* 63). In July, 1947, one year and nine months after insertion, the plate was removed. At operation, all the tissues were very œdematous, pus was present superficial and deep to the plate, which was readily movable in all directions; infection of the bone edges was also present (*Fig.* 64). After removal of bone back to healthy dura, the wound was completely closed; there was a little discharge from the wound in the first few days after operation, but it healed quickly. Full chemotherapy was given. Cultures of the pus grew *Staph. aureus*, relatively insensitive to penicillin. Histological examination of the tissues along the sinus track showed, in addition to inflammatory changes, several foreign-body particles occurring in long spicules which had excited a marked giant-cell reaction; they were not doubly refracting and Major G. B. S. Roberts, R.A.M.C., reported that they were certainly not stone, suture material, or sulphonamide, but most likely bakelite or similar substance.

Why this case became infected is not certain, but it seems that the slipping of the plate was an exciting factor. The foreign particles found later in the wound presumably came from the bomb fragment; it is just possible that their presence was another factor in the case.

Case 5.—*HN*/9376. On Aug. 2, 1944, this patient sustained a compound depressed fracture of the right frontal region, involving the sinuses but without tearing the dura. An extensive removal of the walls of the sinuses and the supra-orbital region was necessary; the mucous membrane of both frontal sinuses was also removed. Six months later a complex tantalum plate was inserted and anchored by pericranial sutures and by two wire sutures

to bone, one to the right external angular process and the other to the glabellar region.

The patient made an uninterrupted convalescence, but seven months after this plastic repair, was bitten by a mosquito just above the right eyebrow over the plate. Three days later a large fluctuant swelling appeared on the forehead. There was no nasal or sinus infection at the time. Aspiration yielded 72 c.c. of pus, which grew a penicillin-sensitive pneumococcus; after three aspirations, combined with local and general penicillin and sulphadiazine, the infection subsided.

A follow-up eighteen months after this episode showed that he was well and the plate satisfactory, but he complained of headache over the right eye.

Cas 6.—*HN*/11721, Nov. 1, 1944. A blunt head injury resulting in a compound depressed fracture of the frontal region and involving both frontal sinuses with

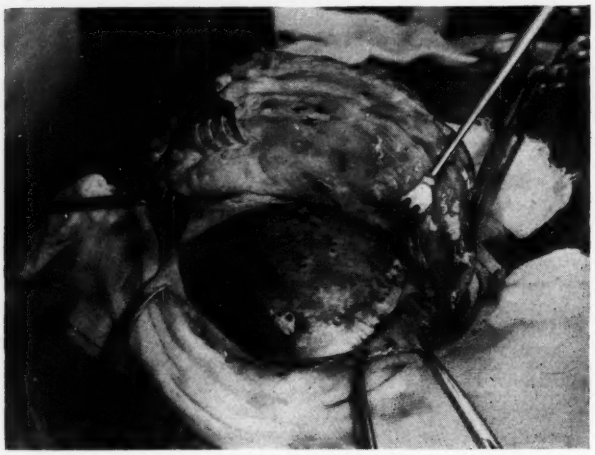

FIG. 65.—*Case* 7. Chronic infection around a tantalum plate. The plate had been inserted partly beneath the pericranium. Note the pyogenic membrane on the deep surface of the œdematous pericranium and on the under surface of the scalp.

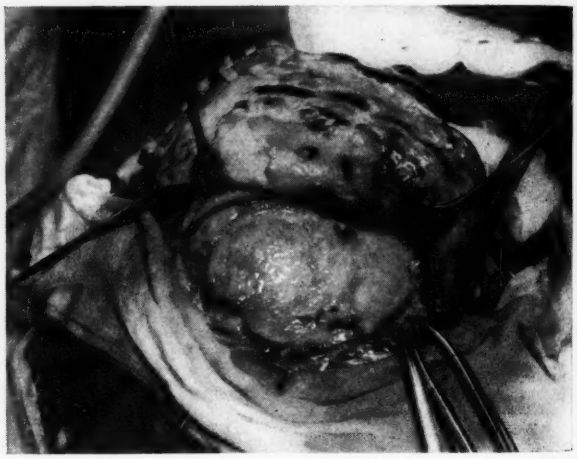

FIG. 66.—*Case* 7. The bony defect is covered by yellow, granulation tissue. There was no pus present and the bony margins seemed healthy.

development of C.S.F. rhinorrhœa. At operation both frontal sinuses were exenterated and a large fascia lata graft placed over the torn dura extradurally. He made an uninterrupted convalescence. Seven months later a tantalum plate was inserted. Again his convalescence was uneventful but the cosmetic result was poor.

He returned to his work as a landscape gardener but thirteen months after the operation he was taken ill with general malaise, headache, and fever. He was admitted to hospital where he was found by Mr. A. MacKenzie Ross to have a bilateral maxillary sinusitis and an abscess forming over the frontal plate. The abscess was drained and culture of the pus grew *B. Friedländer* and diphtheroids; a probe passed through the scalp incision went down to the plate. Both antra contained pus and were washed out. The drain was removed after 48 hours and he made a satisfactory convalescence.

Reporting a year after this episode, the patient said that he was back at work but he was getting pain over the left side of the plate and that it had not felt quite the same since the infection.

Case 7.—*HN*/11600, April 1, 1945. A penetrating right frontoparietal wound which, at the débridement sixty hours after wounding, was already infected and discharging pus, which on culture grew non-hæmolytic streptococci and *B. coli*. The patient developed a post-operative brain fungus which twelve days later was excised and the wound closed except for a small part posteriorly. The scalp finally healed two months after wounding. There were no residual intracranial foreign bodies. Thereafter he made satisfactory progress but for a Jacksonian fit three months after wounding.

A tantalum repair was carried out two months after the scalp had healed. Apart from another fit on the day following operation, he made a good convalescence and the wound healed by first intention.

He reported two years after the cranioplasty that he had continued to have fits at about two-monthly intervals, and in September, 1947, had had four in rapid succession. In addition, fluid had collected over the plate on three occasions; he ascribed the first two attacks to chewing forcibly; there was no pain or tenderness and the fluid disappeared after a fortnight. The third attack, however, coincided with the series of fits in September and this time the fluid took seven weeks to disperse.

As a consequence of this report, the patient was readmitted to hospital in November, 1947. By that time no fluid could be demonstrated beneath the scalp but the plate was loose and no longer fitted well. A lumbar encephalogram showed a traction displacement of the ventricular system towards the site of the defect; the C.S.F. was normal. He was re-explored through a right frontal flap with the object of excising the cortical scar to diminish the frequency of the epileptic attacks, and adjusting the plate.

After reflection of the scalp flap, it was clear that the tantalum plate was the site of a low-grade infection. Turbid yellow fluid was seen coming from beneath the plate, the pericranium was œdematous, and a thin yellow pyogenic membrane, adherent to the deep surface of the scalp, had covered the plate (*Fig.* 65). The plate was freely movable and no suture material to the pericranium could be identified; after removing the plate, a layer of yellow granulation tissue covering the defect was seen, but no pus and no obvious infection of the bony edges (*Fig.* 66). In view of these findings, nothing further was done and the wound was closed after dusting with penicillin-sulphamezathine powder, 5000 units/gramme. A slight growth of paracolon bacteria was obtained on culture of the fluid from beneath the plate and culture of the granulation tissue grew *Staph. albus.*

The history of this case suggested a simple serous collection of fluid from irritation of the plate in relation to the temporalis muscle but at operation there was no doubt that a low-grade infection was present. The tantalum repair had been done only two months after healing of a badly infected brain

fungus, and it may be assumed that the infection was present throughout the twenty-seven months the plate was in, and indeed would not have been disclosed then had it not been for the increasing epileptic attacks.

In the light of these findings it seems very probable that the following 2 cases are also examples of low-grade infection which have subsided without treatment.

Case 8.—*HN*/10034, Nov. 30, 1944. Non-penetrating missile wound of the left frontal region involving the supra-orbital ridge and frontal sinus, with destruction of the left eye. The wound healed by first intention and three months later a tantalum plate was inserted.

He reported two years and seven months after his cranioplasty that the plate was comfortable and cosmetically satisfactory, but on three occasions he had had a collection of fluid over it which each time had settled spontaneously within a week.

Case 9.—*HN*/10082, Oct. 4, 1944. Penetrating missile wound, right frontal region not involving the sinus. There were no complications and tantalum repair was carried out two and a half months later.

On his progress report, the patient's doctor stated that two years and four months after the cranioplasty, a crop of vesicles appeared over the plate and two months after that the right eye became œdematous; both conditions subsided without treatment.

These cases illustrate the ways in which plates can give rise to infection:—

1. An inadequate scalp or conjunctival cover over the plate may result in pressure necrosis and infection (*Case* 1).

2. Although our results show that repair may be safely carried out three months after healing of an infected wound, our experience in *Case* 7 suggests that to attempt it at an earlier date is unwise.

3. Inadequate fixation of the plate with irritation of the tissues by constant movement sets up local conditions favourable to infection (*Case* 4).

4. However inert tantalum may be in the tissues, it still remains a foreign body and in the event of local injury or superficial infection may interfere with the response of the natural defences of the body (*Cases* 3, 5).

5. The relationship of the plate to the frontal sinus (*see below*).

Frontal Sinus Wounds.—Five of the 9 cases described above had wounds involving the frontal sinus. Does this site carry a special liability to infection?

In this series there were 33 frontal sinus wounds. Where the dura was torn in relation to the sinus a fascial repair was carried out, sometimes at the primary operation, but more often within the first few weeks after the original wound was soundly healed, the tantalum plate being inserted about six weeks after that; in 10 cases, however, the dural tear was repaired at the time of cranioplasty. The frontal sinus had been exenterated in some cases, but where the sinus was still patent at the time of cranioplasty a barrier of fascia lata, pericranium, or frontalis muscle was usually interposed between the sinus and the cranial cavity. Cranioplasty was performed earlier in this group of cases than in the series as a whole since a dural repair was often urgently required on account of rhinorrhœa or the risk of meningitis. Sixteen cases had their repair within four months of wounding.

Two of these 33 patients are untraced; the follow-up of the remaining 31 shows that only 16 have reported the plate as entirely satisfactory in appearance and comfort. One patient developed a post-operative meningitis and a second died a year later in status epilepticus with symptoms suggestive of meningitis; 4 became frankly infected and a fifth case has symptoms suggestive of minor infection; 4 patients were dissatisfied with the cosmetic result; and 4 patients have complaints of minor discomfort. The incidence of complications was evenly distributed between those repaired within four months, and those at a later date after wounding.

In assessing these results one must differentiate between those complications arising from the position of the plate in relation to the sinus and those by virtue of the defect being frontal. Reviewing the case histories of those with infective complications, 3 (*Cases* 2, 6, 8) can be attributed directly to the plate-sinus relationship. Plating in relationship to the frontal sinus carried, then, an additional risk of sepsis of 9 per cent. Perhaps this incidence could be lowered by ensuring, with greater care than was always exercised in this series, that the plate was not in open contact with the sinus: but it is fair to say that whether a foreign substance or autogenous bone is used, cranioplasty of this region will always be attended by an increased risk of infection compared with the rest of the skull.

Optimum Time for Tantalum Repair.—How soon can one repair defects after wounding without risk of post-operative infection? We have had no experience of immediate tantalum repair, but the results so far of those cases repaired three months after wounding are favourable.

We have shown that cranioplasty performed three months after healing of an infected wound carries no additional risk; in a small series where a cerebral abscess or its scar was excised at the same time, or the operation was performed shortly after the subsidence of a meningitis, the results are similar. The incidence of complications in 9 cases of frontal sinus wounds repaired three months after wounding was no greater than in the group as a whole. Finally, of 29 cases in the whole series repaired within three months of injury, only 1 case became frankly infected.

We conclude, therefore, that in this series the incidence of infective complications referable to the tantalum plate was 7 per cent; insertion of the plates in relationship to the frontal sinus carried an additional risk of 9 per cent. The results suggest that there is no additional risk in plating three months after wounding or after healing of an infected wound.

INTRACEREBRAL COMPLICATIONS

If an intracranial operation has to be performed some time after the cranioplasty, the tantalum may be left undisturbed and turned down with the bone-flap, as was done successfully in one case of this series where excision of a cerebral scar for epilepsy was performed. On the other hand, if further complications indicate air studies, then the plate, since it is radio-opaque, may so obscure the ventricular outlines that preliminary removal is required; or removal may be required to allow a direct approach to an underlying cerebral lesion. In one case it

was removed two years after insertion on the supposition that it was possibly the cause of the patient's febrile symptoms; the plate was perfectly satisfactory and a ventriculogram disclosed a space-occupying lesion subsequently shown to be an abscess.

It is rather surprising to note that 3 of the 4 plates removed for intracerebral symptoms were in cases with localized hydrocephalus or a condition closely allied to it, as described by Cairns, Johnson, Northcroft, and Daniel (1947). All three had certain features in common: they were penetrating parietal wounds due to missiles and healed by first intention without complications; the protein in the lumbar C.S.F. remained persistently high up to 100 mg. per cent five to fourteen months after wounding; when the tantalum plate was inserted the dura was nicked, with escape of C.S.F., although in one case a pericranial graft was sutured over the rent.

The primary pathology is either an obstruction to part of the ventricular body with steady enlargement of the temporal horn by C.S.F. from its contained choroid plexus, or a cyst communicating with the ventricle by some sort of valve mechanism. The result is the same—a large C.S.F.-containing cyst which acts as a space-occupying lesion and which may be treated by emptying the cyst and coagulating or excising the corresponding portion of the choroid plexus.

In these cases, the ready escape of C.S.F. when the dura was torn, and the fact that in one case where a preliminary air encephalogram was done before the cranioplasty a large porencephalic cyst was demonstrated beneath the defect, make it a reasonable assumption that the cerebral lesion was present at the time of operation. But does tantalum in contact with the ventricular system stimulate further C.S.F. secretion?

In the first case the symptoms of increased pressure did not arise for 15 months after the cranioplasty and then excision of the choroid plexus resulted in rapid subsidence of symptoms; the plate which had been removed nine days previously was replaced at this second operation and one year later was satisfactory.

The second case, however, had a persistent collection of subgaleal C.S.F. for six weeks after the repair. This settled down, but two months later the patient suddenly developed status epilepticus. The plate was riding and, as the status continued, the wound was re-opened; C.S.F. poured out. The plate was removed and it was seen that most of the hemisphere was replaced by what appeared to be ventricle. He died very shortly after operation; there was no autopsy.

In the third, the relationship between onset of symptoms and cranioplasty was immediate. The patient was making a satisfactory recovery from his head wound until he had the tantalum repair. Following operation he had a large sub-galeal collection of C.S.F. which for fifteen months persistently recurred despite all conservative measures, including repeated aspirations, lumbar punctures, and pressure dressings; he finally developed symptoms of increased pressure with papilloedema and was re-explored. The tantalum was in direct contact with a large intracerebral cyst over an area about 1 cm. diameter, despite the fact that a pericranial graft had been sewn over the dura at the time of cranioplasty. Choroid plexus was seen in the wall of the cyst; this was coagulated and the plate removed, with rapid clearing of his symptoms.

It may be that localized hydrocephalus after parietal wounds is far commoner than supposed, yet in view of these experiences within a relatively small series of cases we cannot dismiss the possibility of tantalum as an exciting factor. Investigation into the reaction of tantalum in contact with the ventricle might yield further information.

EPILEPSY

So far, 51 of these patients have had fits. Whether repair of a skull defect with tantalum affects the incidence of epilepsy or frequency of attacks is difficult to evaluate correctly since so many of these cases were repaired a few months after wounding or before one could estimate the likely frequency of the attacks; moreover, all these patients were given anticonvulsant treatment, usually phenobarbital. Late results must be offset against the natural course of traumatic epilepsy without skull repair. Thus the majority of patients will have their first fit within eighteen months of wounding (Russell, 1947); Ascroft (1941) observed that about one-third of patients with traumatic epilepsy suffered for the first few years only and the attacks then ceased, a very important factor in assessing the value of an operation such as cranioplasty. The effect of excision or removal of foreign bodies at the time of the repair must also be considered; such an operation was performed in 13 of these cases.

Ten of these 51 patients may be excluded from the discussion; 7 only had fits within the first month of wounding, in 2 the only fits followed within a fortnight after the cranioplasty; 1 patient was untraced. There were 20 patients who had fits before the operation and 21 whose first fit occurred some time after the operation.

Of the 20 patients who had fits before the operation, 9 had an intracerebral operation at the time the plate was inserted—excision of brain scar, removal of bone-chips, excision of cerebral abscess—and the late results showed that this group did well; 3 have had no further fits; 4 are improved and 2 unchanged. In 11 patients where the operation consisted of skull repair only, the frequency of the fits remained about the same, two years or more later, in 8, and became worse in 3.

Twenty-one patients had their first fit after the cranioplasty. In 8 this occurred within three months of the operation, but in 6 not until over a year later; the longest interval was twenty-six months. The wide variation in the time of onset after plating and the fact that 14 had their first fit within eighteen months of wounding suggest that these patients would have developed epilepsy anyway, and that it is most improbable that the cranioplasty was a determining factor.

Our results up to two years, therefore, do not agree with the findings of Grant and Norcross (1939) that cranioplasty alone benefits a proportion of epileptics. The only patients to improve in this series were those in whom an intracerebral operation had been performed as well; no patient with a repair only has

shown any marked decrease in the frequency of the fits.

Although many of the cases developed their first fit after the operation, consideration of the time after wounding and the interval between the repair and the onset of fits suggests that it is most unlikely that there is any direct relationship. However, the operation may precipitate an attack in an epileptic; as seen from *Table II*, 10 patients had a fit within a fortnight post-operatively, 4 of these within twenty-four hours of the operation despite sedation. For this reason, all cases should have anticonvulsants over the period of operation and probably for at least a fortnight afterwards.

Electro-encephalogram.—It is not proposed to discuss here the relationship between E.E.G. recordings and traumatic epilepsy save to say that the findings were sometimes at variance with the clinical history, and frankly epileptic records or epileptic changes in the E.E.G. did not affect the selection of cases for repair.

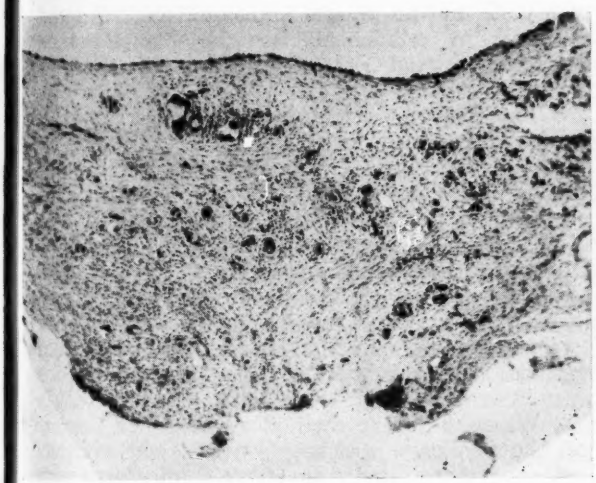

FIG. 67.—Thin membrane formed around a tantalum plate. Note the loose collagenous tissue, numerous small blood-vessels and a single layer of lining cells. Hæmatoxylin and van Giesen. (× ½.)

The presence of a skull defect or tantalum plate affects the character of the E.E.G. recording and Dr. Denis Williams, who was responsible for the E.E.G. recordings in this series, has kindly written the following comment:—

"The electro-encephalographic tracings which are obtained from the scalp are usually seen equally well on the right and left sides so long as no lesion exists in either cerebral hemisphere. The symmetry of the recordings is in part dependent upon the resistance of the tissues between the brain and the recording electrodes. In abnormal states, the physical properties of the skull and scalp may be disturbed, and in particular the disturbance may be associated with an alteration in the electrical resistance of the tissues. This alteration may be in the direction of a decrease or an increase. For instance, in a vascular scalp or in a state of activity of the sweat glands, the resistance is reduced, whereas with an atrophic or dry scalp, the resistance is high. Conditions of the bone may similarly affect the conduction between the brain and the electrodes. It is a common finding that even in the presence of a linear crack, and much more so with a single burr-hole, the amplitude of the rhythms recorded on the affected side is greater than on the other. When large bone defects are present, the change is even more striking.

"When a bone defect has been repaired, the effect on the E.E.G. records depends upon the nature of the filling material. It also depends upon whether one or more of the recording electrodes which are used in the input of any recording channel lie over the graft. Metallic plates such as tantalum increase the conductivity so that if one electrode lies over the plate and one is over normal bone, the plate acts as an enormous electrode and increases the voltage of the rhythms. If, however, the two electrodes are over the tantalum, it is clear that the potential changes short-circuit through the plate, with resulting reduction in the size of the recorded electrical signals. When, on the other hand, a non-conducting medium such as acrylic is used, there is a reduction in the amplitude of the signals whether one or both of the electrodes of the recording system lie over the plate. A careful evaluation of the physical situation is therefore essential whenever electro-encephalograms are made in patients who have had a penetrating head injury."

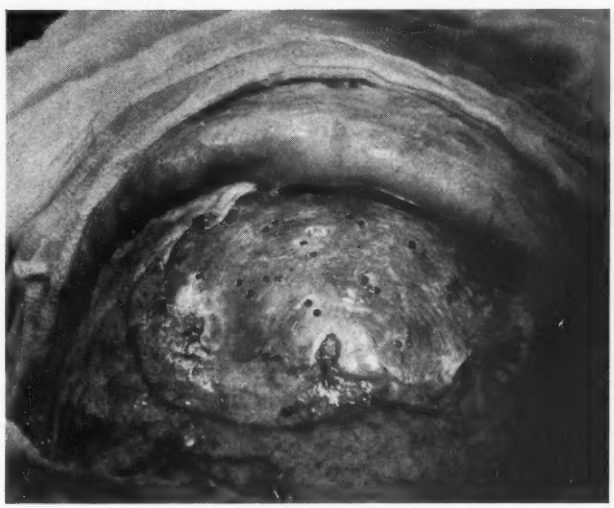

FIG. 68.—Fibrous tissue growing through the holes in the plate. Appearance at operation.

TISSUE REACTION TO TANTALUM

Observations on cases where plates have been removed for non-infective complications confirm the early promise of the experimental work that the metal is surprisingly inert in the tissues. A minimal fibrous tissue reaction is excited with the formation of a thin membrane on each side of the plate so that it ultimately becomes embedded in a fibrous tissue sac. In one case of this series where the tantalum plate was included in a bone-flap for a further operation eighteen months after its insertion, Mr. J. Pennybacker reported that the galea stripped easily from the plate; it was enclosed in a thin translucent membrane of about the same thickness as pericranium and the perforations of the plate were visible through it; the bone edges

were smooth and round and the impression given was that a minimal reaction had occurred sufficient only to keep the plate securely in place. Histological examination of the membrane from another case showed it to consist of loose collagenous tissue containing numerous small blood-vessels with some cuffing by chronic inflammatory cells, and foreign-body giant cells; there was a single layer of lining cells along the surface in contact with the plate (*Fig.* 67). The membrane is similar to that formed around acrylic resin (Beck, Russell, Small, and Graham, 1945). In some cases the fibrous tissue may grow through the perforations in the plate; *Fig.* 68 shows the appearance at operation in such cases, and *Fig.* 69 is an autopsy specimen of a case (not in this series) where this process was very marked and had anchored the plate very securely.

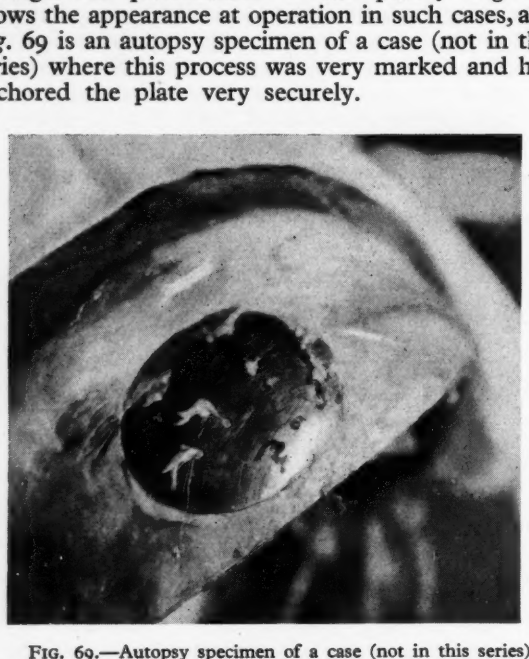

FIG. 69.—Autopsy specimen of a case (not in this series) to show marked fibrous growth through the holes in the plate, thus anchoring the plate very securely.

Where infection occurs, the plates retain their surface and lustre without evidence of corrosion. *Cases* 5 and 6 afford striking evidence of how well the tissues tolerate tantalum; in these cases frank pus formed around the plates but the infection subsided with drainage of the pus and penicillin therapy and without necessitating removal of the plate. *Case* 7 shows how a low-grade infection can exist around a plate for over two years without definite signs.

SUMMARY AND CONCLUSIONS

1. The use of tantalum for repair of traumatic skull defects at the Military Hospital for Head Injuries, Oxford, is described, and the immediate and late results of the first 128 cases followed up for a period of at least two years are recorded. Only four patients, who have gone abroad, are untraced.

2. Of the 124 cases traced 94 have been completely satisfactory and without any complications two years or more following operation. Eleven of the 130 plates inserted have been removed, 5 on account of infection, 4 for an intracerebral complication; 2 were cosmetic failures.

3. There were no post-operative deaths and the convalescence was usually uneventful, but most cases developed a serosanguineous fluid collection beneath the scalp which required aspiration. In 3 cases a subgaleal collection of C.S.F. persisted for several weeks. There was 1 case of post-operative wound infection.

4. The cosmetic results were generally very good. The accurate shaping of complex plates for the frontal defects involving the orbital margin is difficult; 8 of the 68 frontal plates were not entirely satisfactory, although only 3 were definite failures. Defects over the vault were uniformly successful.

5. The incidence of infection, both early and late, was 7 per cent; 5 plates had to be removed on this account. The exciting causes of infection were: poor skin cover; too early plating after wound infection; slipping of the plate; in the region of the plate lack of normal cellular response to injury or potential infection; and the relationship of the plate to the frontal sinus. It is estimated that plates in relationship to the frontal sinus carry an additional risk of infection of about 9 per cent.

6. There was no evidence that cranioplasty with tantalum diminished the incidence or the severity of traumatic epilepsy. On the other hand the operation may precipitate an attack in an epileptic. This occurred in 20 per cent of the epileptic group. For this reason all patients should be given anticonvulsants during the operative period.

7. The results suggest that plating may be successfully carried out three months after wounding or after healing of an infected wound without increase of complications over those in cases repaired after a longer interval of time.

8. When the onlay method as described here is used, the advantage of tantalum over other materials is evident; it is a simple one-stage cranioplasty with little strain on the patient. To obtain good results, however, requires the most careful preparation of the plate and attention to the condition and vascularity of the overlying scalp.

In addition to the frequency of post-operative collections of fluid beneath the scalp, there is a possibility that tantalum in contact with the ventricular system may stimulate the production of C.S.F. For these reasons the dural cover beneath the plate should be complete; all tears should be very carefully sutured or repaired with a fascia lata or pericranial graft.

9. Tantalum is a satisfactory material for cranioplasty of the vault; its main disadvantage is that it is radio-opaque and would not be indicated in those cases where it was thought that air studies might be required in the future.

The results of this first series suggest that it is probably not the method of choice for defects in the low frontal region since the cosmetic result cannot be guaranteed with a one-stage procedure, and the plates tend to slip unless special precautions are taken. An increased risk of infection in those cases involving the frontal sinus is present whatever material is used for plastic repair, even with all the advantages of modern chemotherapy; it is probably less with autogenous bone than with tantalum or, for that matter, acrylic resin, but precise data on this question are still lacking.

Our thanks are due to Dr. W. Ritchie Russell, who gave us every assistance and allowed us the full use of the records and organization of the Head Injury Bureau, Oxford; to the several doctors, in addition to those mentioned in the text, who kindly sent us reports on individual cases; To Mr. E. L. Tugwell and Pte. K. W. Goddard, R.A.M.C., for the photographs; to Mr. L. G. Martin for technical assistance in preparing the tantalum plates; and to Professor Sir Hugh Cairns for his advice in the preparation of this paper.

The success of the follow-up returns of these patients was largely due to the work of the Almoner, Miss D. Martin-Hurst, who had personal knowledge of many of these patients; and through her initiative it was possible to trace patients with the help of the British Red Cross Organization and other agencies whose co-operation we gratefully acknowledge.

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LYMPHADENOID GOITRE

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EVER since Hashimoto (1912) first described struma lymphomatosum, numerous papers, including over 200 case reports dealing with this condition, have appeared. Struma lymphomatosum, lymphadenoid goitre, or Hashimoto's disease, is characterized by a diffuse goitre occurring in women of menopausal age. The thyroid is at first of normal consistency, later becoming firm or hard, but never adherent to its surroundings. The goitre usually causes symptoms by pressure on surrounding structures. Thyrotoxic symptoms are often present at the onset. Later the tendency is towards myxœdema. Histologically a distinct picture is described, in which extensive lymphocyte infiltration has replaced most of the thyroid vesicles and the surviving epithelial cells are often of atypical appearance. The final diagnosis of lymphadenoid goitre is usually made on histological evidence. Round-cell infiltration is, however, met with in a number of conditions of the thyroid gland, including tuberculous thyroiditis, syphilitic thyroiditis, acute thyroiditis, chronic thyroiditis of giant-cell type (De Quervain, 1904), Riedel's thyroiditis, myxœdema, and following prolonged iodine medication. Furthermore, Simmonds (1913) has shown that lymphocytic aggregations and infiltrations occur in 5 per cent of healthy thyroids above 30 years of age, in 15 per cent of cases of non-toxic goitre, and in 75 per cent of toxic goitres. Hellwig (1938) found small islands of tissue histologically indistinguishable from lymphadenoid goitre in 23 per cent of toxic goitres removed surgically.

Thus the histological picture of lymphadenoid goitre depends on the greater extent and intensity of the lymphocytic infiltration to distinguish it from the lesser infiltrations that may be seen in hyperthyroidism, hypothyroidism, and endemic goitre. The distinction appears to be quantitative and arbitrary; the criteria that have been used in the literature vary.

Schilling (1945), for instance, excludes all round-cell infiltration, fibrosis, and lymphoid hyperplasia associated with simple colloid goitre, adenomata, or toxic goitre. On the other hand Eden and Trotter (1942) and Polowe (1934) report cases of lymphadenoid goitre associated with thyrotoxicosis. McSwain and Moore (1943) found basal metabolic rates ranging from +63 per cent to −23 per cent in their series of 71 cases of lymphadenoid goitre. Again, whereas Vaux (1938) described 15 out of her 38 cases as early-stage lymphadenoid goitre, Joll (1939) considered this early stage as merely thyrotoxicosis with associated lymphoid infiltration.

This study is an attempt to find out, first, whether it is possible to differentiate clearly between thyrotoxicosis or simple goitre with lymphocytic infiltration on the one hand and lymphadenoid goitre on the other; and, secondly, to determine whether a study of the relationship between the two conditions can throw any light on the aetiology and development of lymphadenoid goitre.

In the first instance 121 cases were examined which had undergone thyroidectomy for various reasons and in which the histological picture showed varying degrees of round-cell infiltration from multiple lymphocytic foci to the appearances first described by Hashimoto. As a result of our findings we felt it necessary to investigate round-cell infiltration of even slighter degree and a second series of 64 thyroids removed for various causes was studied. In this series cases with prominent lymphocytic infiltration were excluded.

METHOD

First Series.—Paraffin sections of blocks of thyroid tissue were stained with hæmatoxylin and eosin; in 49 cases large sections comprising a part

or whole of a thyroid lobe were available. The following points in each case were noted :—

Age; sex; length of history; absence or presence of a previous goitre; absence or presence of toxic and pressure symptoms; previous treatment; eye signs; type and consistency of goitre; pulse-rate and B.M.R.; basic histological picture, whether toxic, subtoxic, colloid goitre or adenomatous hyperplasia.
Amount of colloid in vesicles.
Amount of lymphoid foci.
Presence or absence of germ centres.
Degree of infiltration of lymphoid tissue.
Atrophy and disorganization of vesicles.
Fibrosis.
Hürthle cells.
Changes in blood-vessels of large and small calibre.

Wherever possible a system of grading + to +++++ was employed.

Second Series.—Large sections comprising part or whole of a thyroid lobe were prepared by the paraffin method and stained by hæmatoxylin and eosin. The following points were noted in each case :—

Basic picture.
Condition of vessels.
Presence or absence of subtoxic vesicles and lymphoid foci with special attention to their location.

RESULTS

FIRST SERIES

HISTOLOGICAL

The 121 cases were divided into the following six classes on histological grounds :—

1. Incipient lymphocytic infiltration	9
2. Minimal lymphocytic infiltration	27
3. Moderate lymphocytic infiltration	37
4. Marked lymphocytic infiltration	25
5. Fully developed Hashimoto's disease ..	10
6. Atypical histological picture	13
	121

The staging was based on the amount of lymphoid infiltration and on the associated changes in the thyroid epithelium.

1. Incipient Lymphocytic Infiltration.—Scattered lymphoid foci with slight infiltration of round cells between vesicles, which may show early atrophic changes (*Fig.* 70).

2. Minimal Lymphocytic Infiltration.—Lymphoid foci are more numerous and often larger. Round-cell infiltration between vesicles is more prominent and small areas of atrophic and/or disorganized vesicles may be seen in relation to the infiltrating cells (*Fig.* 71).

3. Moderate Lymphocytic Infiltration.—Large areas of thyroid tissue contain lymphoid foci. Infiltration is widespread among vesicles, a considerable number of which show changes of atrophy and disorganization. Whole areas of thyroid tissue are still unaffected.

4. Marked Lymphocytic Infiltration.—Almost all fields show lymphadenoid changes, but a considerable number of normal thyroid vesicles can still be seen.

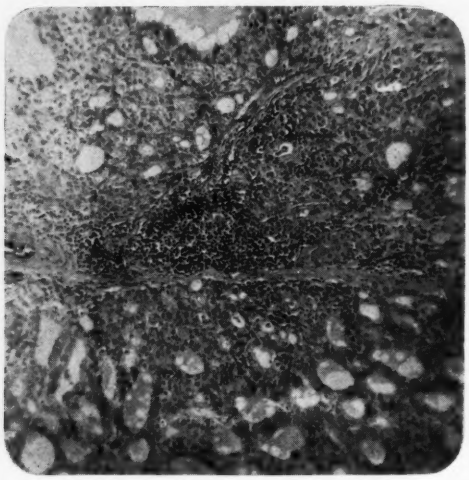

FIG. 70.—Exophthalmic goitre with a lymphoid focus. H. and E. (× 76.)

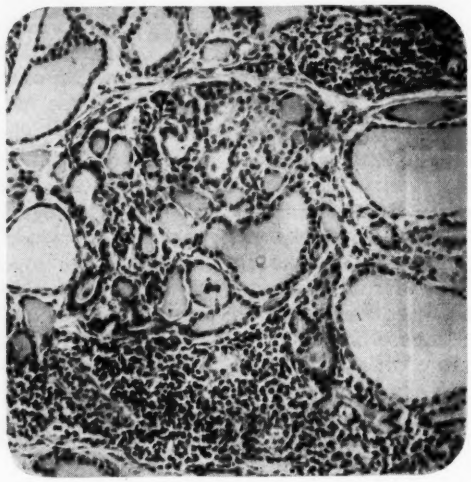

FIG. 71.—Lymphocytic infiltration with atrophy of vesicles. H. and E. (× 136.)

5. Hashimoto's Disease.—Extensive lymphoid infiltration with but few surviving groups of atrophic and disorganized vesicles still recognizable. The classical picture (*Fig.* 72).

6. Atypical Histological Picture.—Thyroid epithelium shows atypical cytology and arrangement, lymphoid infiltration variable, a definite diagnosis of lymphadenoid goitre cannot be made.

The individual histological findings can now be considered in detail.

1. Lymphoid Tissue.—Lymphocytes occurred in rounded masses in lymph-follicles, or as diffuse infiltrations in the thyroid tissue. The *follicles* varied in size and number and in the fully developed stages no obvious anatomical location could be recognized. In the less advanced stages they were usually seen in greater numbers towards the periphery of the gland. In glands showing lobulation they tended to be located in the perilobular connective tissue. Where distinct areas of adenomatous

hyperplasia were present, lymphadenoid changes were frequently seen in the thyroid tissue immediately adjacent to the adenomata. Adenomata themselves were always free from lymphocytic infiltrations.

Germ centres in the lymphoid follicles were present in all our cases, except 1 in the incipient, 8 in the minimal, 6 in the moderate, 4 in the marked, and 1 in the advanced groups. Joll (1939) and Schilling (1945) consider the presence of germ centres characteristic of the disease, but their presence in even the earliest stages of our series seems to indicate that they are of no more significance than the lymph-follicles themselves.

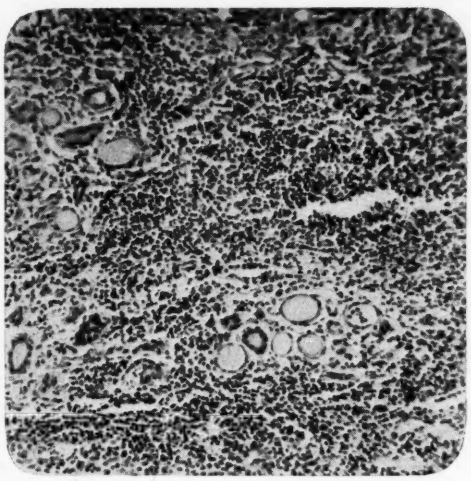

FIG. 72.—A field from a fully developed Hashimoto's disease. H. and E. (× 120.)

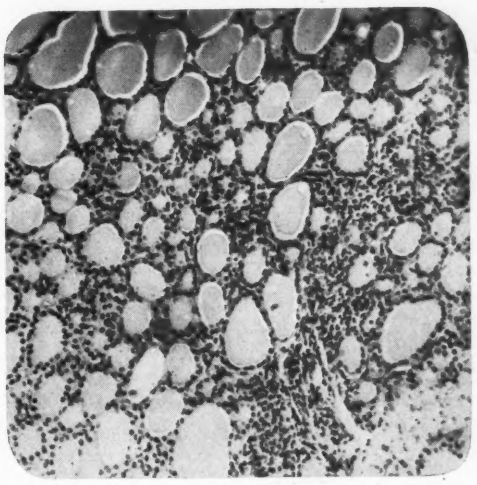

FIG. 73.—Subtoxic vesicles with slight lymphoid infiltration. H. and E. (× 120.)

2. Diffuse Lymphoid Infiltration.—Here the lymphoid cells are present in small groups between the acini. Usually the extent of the infiltration was proportionate to the size and number of the lymphoid follicles. There was a close association between the amounts of diffuse lymphoid infiltration and atrophy, as well as disorganization of the thyroid vesicles.

3. Fibrosis.—Fibrosis of the inter-acinar connective tissue was absent or minimal in the earlier groups and more pronounced and widespread in the later ones. Similar findings are described by Vaux (1938), who advanced this fact in support of the view that lymphadenoid goitre is an early stage of Riedel's thyroiditis.

4. Epithelium.—The basic picture of the thyroid epithelium was variable. Sometimes it was uniformly that of exophthalmic goitre (*Fig.* 70), sometimes predominantly that of colloid goitre. At other times it was mixed. In the fully developed Hashimoto's disease, the surviving epithelial cells were quite atypical. In many cases, however, the majority of the vesicles tended to be small and of similar size. The epithelium was mainly flattened or cubical (*Fig.* 73). This type of vesicle we have described as subtoxic, following Vaux, and, as may be seen from *Table I*, it was the most frequently occurring basic histological picture. It was commoner in the more advanced group (38 out of 75) than in the earlier ones (12 out of 35). At the same time the background picture of exophthalmic goitre showed an opposite tendency, being commoner in the earlier groups (13 out of 35) than in the more advanced groups (14 out of 75).

5. Colloid.—Colloid was variable. It was often normal in amount. In the later stages it was usually considerably diminished.

6. Atrophy of Vesicles.—Closely associated with lymphoid follicles and diffuse lymphoid infiltration were vesicles of small size and lacking colloid. The epithelium was cuboidal and the cell nuclei were hyperchromatic (*Fig.* 74). These changes were interpreted as representing atrophy, though it is not certain that these small acini are in the process of disappearing. Later stages, however, show a progressive increase in the number of these vesicles in conjunction with increase in lymphoid infiltration. Later still the acinar structure becomes lost in a mass of lymphoid cells.

7. Hürthle Cells.—Groups of epithelial cells usually forming small ill-formed acini without colloid and possessing much eosinophilic cytoplasm were present in about half of the cases. The nuclei tended to be large, vacuolated, and hyperchromatic. Such cell groups were characteristically embedded in lymphoid masses or closely associated with lymphoid foci (*Fig.* 75). In 11 cases, classed in the atypical group, these cells were present in large numbers and not associated with lymphoid foci. In 3 of

Table I.—INCIDENCE OF TYPES OF HISTOLOGICAL BACKGROUND

STAGE	HISTOLOGICAL BACKGROUND					
	Toxic	Subtoxic	Mixed toxic and Subtoxic	Normal	Adenoma	No Pattern Visible
Incipient	2	2	3	1	1	–
Minimal	11	10	4	–	1	–
Moderate	7	20	9	1	–	–
Marked	6	13	4	–	1	–
Fully developed	1	5	–	–	1	3

these cases definite adenomatous masses of these cells were present.

The cells seem to correspond to the cells rich in protoplasm of Hürthle (1894). In our cases they were about equally frequent at all stages of lymphoid infiltration (*Table II*).

Table II.—Showing Incidence of Hürthle Cells

Hürthle cells	*Incipient*	*Minimal*	*Moderate*	*Marked*	*Full*	*Total*
Present	6	15	15	14	6	56
Absent	3	12	22	12	4	53

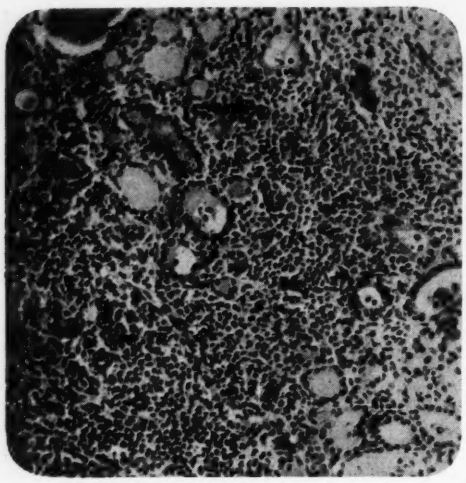

Fig. 74.—Atrophic vesicles with intense lymphocytic infiltration. H. and E. (× 120.)

8. Giant Cells.—Multinucleate giant cells of Langhans type were only an occasional finding and were present when disorganization and fibrosis were marked features.

9. Blood-vessels.—The smaller and medium-sized arteries penetrating the thyroid substance showed no definite pathological changes. In 49 out of 121 cases, sections of the larger arteries which course over the surface of the gland were available. Pathological changes were seen in 19. The commonest change was widespread calcification of the internal elastic lamella; more rarely intimal hyperplasia was seen.

The average age of the cases showing vascular changes was 50 years, and of those without changes 41 years. The average length of history in cases with arterial change was 8·8 years; without, 3·5 years. The vascular changes showed no relation to the degree of lymphocytic infiltration.

Clinical

Sex.—Only 2 of the 121 cases were males. One of these had moderate lymphocytic infiltration and the other a generalized Hürthle cell change. This sex incidence corresponds to that generally found in Hashimoto's disease.

Age.—The ages ranged from 15 to 66 years.

Table III.—Age in Relation to Lymphocytic Infiltration

	Incipient	*Minimal*	*Moderate*	*Marked*	*Fully developed*
Average age	41	43·7	45	39	47

As can be seen from *Table III* there is a progressive increase in average age with increased lymphocytic infiltration in the first three stages, but in the group with marked infiltration the average age is lower than in the preceding stages. Vaux (1938) notes a progressive increase in age incidence with increasing severity of lymphadenoid changes, but our figures do not fully support this relationship.

History.—The presence of a goitre some years prior to the appearance of other symptoms was recorded in 40·7 per cent of cases.

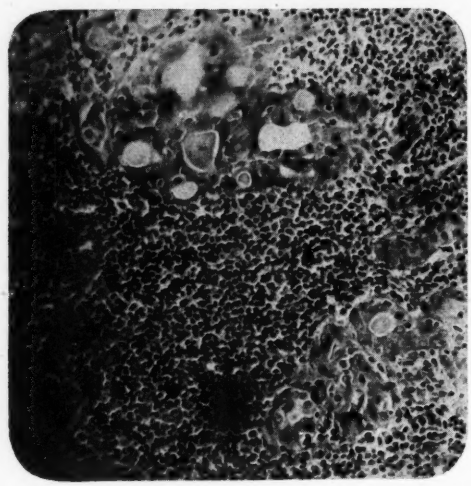

Fig. 75.—Groups of Hürthle cells embedded in lymphoid tissue. H. and E. (× 148.)

Toxic Symptoms.—These were associated with almost all the cases in the groups with incipient, minimal, moderate, and marked lymphocytic infiltrations, and in 4 out of 9 of the fully developed lymphadenoid goitre group. In these 4 cases the mean B.M.R. was raised above 20 per cent. In one of them it was +52 per cent. The mean B.M.R. was highest in those cases which showed generalized Hürthle cell change and lowest in the fully developed group (*Table IV*).

Table IV.—Incidence of Toxic Symptoms

	Mean B.M.R.	Weight		Pressure Symptoms	
		Loss	Steady or Increase	Present	Absent
	per cent				
Minimal L.I.	+ 38	16	8	2	23
Moderate L.I.	+ 30	21	13	5	28
Marked L.I.	+ 33	9	11	5	17
Fully developed L.G.	+ 18	4	5	7	2
Generalized Hürthle cell change	+ 40	16	—	4	12

Four clinical types could be recognized in this series: (1) Primary Graves' disease; (2) Secondary Graves' disease; (3) History of thyrotoxicosis for years, with recent increase in size of goitre; (4) Long-standing Graves' disease treated medically, with recent recrudescence of symptoms. (*Table V.*)

Pressure Symptoms.—The incidence of pressure symptoms increased with each further stage of lymphoid infiltration. The symptoms complained

of were dyspnœa, a feeling of constriction in the neck, and hoarseness.

Physical Signs.—*See Table VI.*

In the later stages the proportion of firm glands is increased. Firmness of the glands appears to be associated with fibrosis rather than with lymphoid infiltration. The thyroid always moved well on swallowing and in no case were adhesions found to surrounding muscles and vessels. No recurrent laryngeal palsy has been observed. The trachea was often narrowed by lateral compression.

Table V.—INCIDENCE OF CLINICAL TYPES

	1. Primary Graves' Disease	2. Secondary Graves' Disease	3. Thyrotoxicosis for Years, Goitre Growth Recently	4. Long-standing Graves' Disease with Recrudescence	5. No Toxic Symptoms
Minimal L.I.	13	2	1	5	–
Moderate L.I.	11	11	1	6	3
Marked L.I.	8	6	3	5	2
Fully developed L.G.	1	3	4	–	2
Generalized Hürthle cell change	6	4	1	5	–

Table VI.—PHYSICAL SIGNS

	Goitre		Consistency		Eye Signs	
	Diffuse	Adenomatous	Firm	Soft	Present	Absent
Incipient L.I.	5	2	–	7	5	2
Minimal L.I.	20	4	5	17	12	12
Moderate L.I.	27	4	3	25	23	11
Marked L.I.	17	5	8	14	8	14
Developed L.G.	7	2	5	4	1	8
Generalized Hürthle cell change	12	2	4	10	12	4

Table VII.—INCIDENCE OF SUBTOXIC VESICLES AND LYMPHOID TISSUE

	Basic Picture No.	Subtoxic Epithelium under Capsule	Subtoxic Epith. and R.C.I. under Capsule	Hürthle Cells	R.C.I. Alone
Adenomatous goitre	42	41	25	5	–
Toxic goitre	16	11	5	1	4
Colloid goitre	6	5	1	–	–

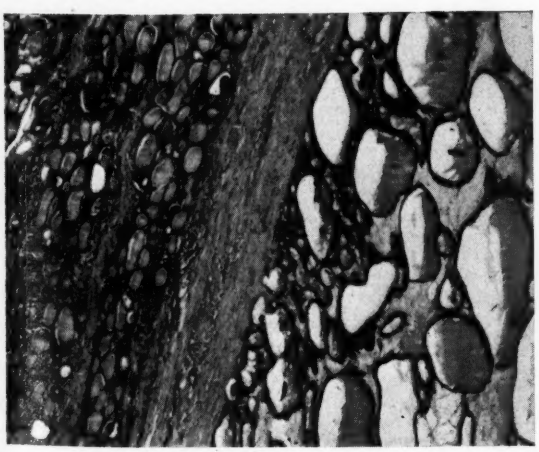

FIG. 76.—The large vesicles of an area of adenomatous hyperplasia (right) are separated by the fibrous tissue capsule from a rim of subtoxic vesicles (left) present in the adjacent non-adenomatous thyroid tissue. H. and E. (× 42.)

SECOND SERIES

(Sixty-four thyroidectomy cases in which lymphoid infiltration was slight or absent.)

Vessels.—In 45 cases large vessels were seen on the surface of the thyroid gland. Of these, 16 showed calcification of the internal elastic lamella or intimal sclerosis. This is a similar incidence to that in the first series, 19 out of 49 cases. The vascular changes therefore are of no significance in lymphocytic infiltration, since they are about as frequent in the first series as in the second series.

Subtoxic Vesicles and Lymphoid Tissue (*see Table VII*).—There were 42 cases of adenomatous goitre. Of these, 41 had areas of subtoxic epithelium in the rim of thyroid tissue (*Fig.* 76) surrounding the adenomata and 8 showed these changes in the thyroid tissue intervening between adenomata. In 25 of these sections lymphoid infiltration was associated with the subtoxic vesicles (*Fig.* 77). In 5 cases Hürthle cells were associated with the round-cell infiltration (*Fig.* 78). Of 16 cases of exophthalmic and toxic goitre, 11 showed subtoxic areas with round-cell infiltration under the capsule. Of these 11 cases 5 also had round-cell infiltration in the substance of the gland. One case showed Hürthle cells. There were 6 cases of colloid goitre; 5 of these presented subtoxic areas under the capsule, one of which also had associated round-cell infiltration and one also subtoxic areas in its substance.

DISCUSSION

Joll's criticism of Vaux's inclusion of her early cases under the term lymphadenoid goitre if valid would apply equally to our first series of cases. The basis of our series is round-cell infiltration and we meet undoubted toxic goitre at one end of the scale and undoubted Hashimoto's disease at the other. All changes present in a diffuse form in Hashimoto's disease are met with focally in a number of cases of toxic goitre. Even the germ centres and Hürthle cells deemed characteristic of lymphadenoid goitre by Joll and others are there. Round-cell infiltration, subtoxic vesicles, atrophy and degeneration of the thyroid epithelium are present in the earliest stages, and with any increase in round cells these features become more marked. An increase in fibrosis as well can be seen with each further stage, indicating a greater extent or a longer duration of the disease process. Clinically there is a gradual decline in toxicity with each stage, but even 4 out of the 10 cases in which round-cell infiltration was extreme had B.M.R.s of +20 per cent or above at the time of the operation and presented with toxic symptoms.

So the evidence suggests that we are dealing with a true sequence and it supports the suggestion made by Eden and Trotter (1942) that lymphadenoid goitre is an exaggeration of the lymphoid infiltration that may occur in toxic goitre.

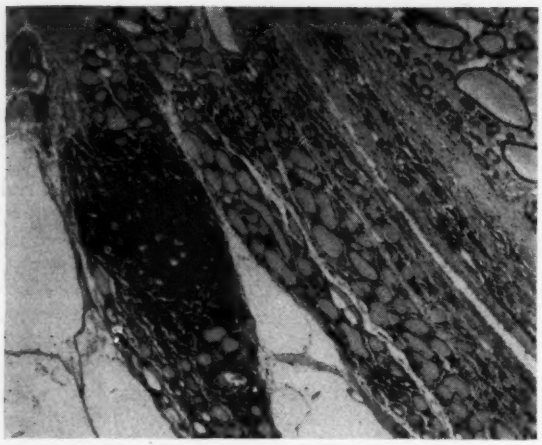

FIG. 77.—Lymphoid focus and subtoxic vesicles (left) separated by a fibrous tissue zone from an area of adenomatous hyperplasia (top right). H. and E. (× 42.)

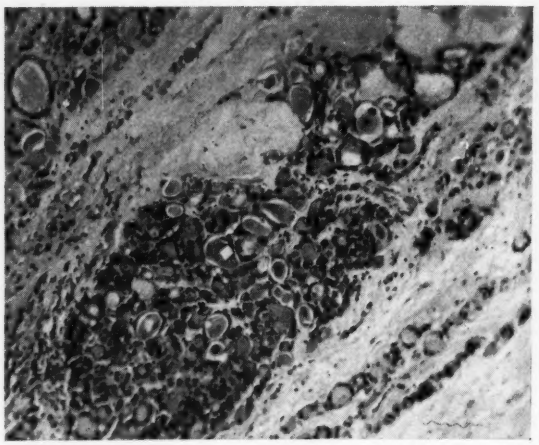

FIG. 78.—An area of Hürthle cells embedded in the fibrous tissue capsule of an area of adenomatous hyperplasia. H. and E. (× 82.)

Most pathological conditions of the thyroid are thought to appear initially as multi-focal processes. This is believed to be so in thyrotoxicosis (Holst and Klose, 1943) and in involution of the gland (Rienhoff, 1925). The above conception of the development of lymphadenoid goitre would bring it into line with that of thyrotoxicosis and degeneration of the thyroid gland.

Connexion between Lymphadenoid Goitre and Riedel's Thyroiditis.—Fibrosis was slight in the less advanced group and was progressively increased in the more advanced stages. Vaux reported similar findings and used these as evidence that lymphadenoid goitre is an early stage of Riedel's thyroiditis, an opinion also held by Ewing (1922) and Heyd (1929). No case of lymphadenoid goitre has ever been observed to change to Riedel's thyroiditis. Histological sections of a recurrent lymphadenoid goitre, removed nine years after the first thyroidectomy, showed no progression towards Riedel's thyroiditis (McClintock and Wright, 1937). The two diseases have been clearly separated on clinical grounds by Lee (1935), McClintock and Wright (1937), Joll (1939), Harvey (1940), McSwain and Moore (1943), and Schilling (1945). Nor has this series succeeded in furnishing evidence that lymphadenoid goitre progresses to Riedel's thyroiditis.

Hürthle Cells.—These cells have been described in association with lymphadenoid goitre by McClintock and Wright (1937), Hellwig (1938), Joll (1939), Womack (1944), and Palmley and Hellwig (1946). They have occasionally been found in Riedel's thryoiditis, and in myxœdema. Twenty-one cases of Hürthle cell tumours have been reported (Morrow, 1945), the majority of which occurred in women over the age of 40; 8 cases were diagnosed as carcinoma, but since none of them recurred the malignant nature of these tumours is doubtful. Various origins of the Hürthle cells have been conjectured. Getzower (1907) suggested that they derived from the post-branchial bodies, Eisenberg and Wallerstein (1932) that they come from the parathyroids, Wilenski and Kaufman (1938) and Schilling (1945) that they arise from normal thyroid epithelium as a result of metabolic, nutritional, or chemical disturbance; Palmley and Hellwig (1946) thought that they represent a compensatory effort following exhaustion of the thyroid colloid. Brünger (1915) and Joll (1939) regarded them as regenerative or hypertrophic cells. Reist (1922), Graham (1931), and Schilling (1945) described them as inactive or degenerative cells.

In those of our cases which showed a generalized change of large parts of thyroid tissue into Hürthle cells, the history of symptoms was relatively short, with an average of 8 months, and the toxicity was marked, averaging a B.M.R. of + 40 per cent. These cells seem to represent hyperactive epithelium in a part of the thyroid which for some reason cannot respond to the thyrotropic stimulus in the usual way.

Subtoxic Vesicles.—These small vesicles were found to be the most frequently occurring background histological picture in the first series and were most frequently found in the groups showing the more advanced stages of lymphocytic infiltration. Irrespective of the state of the rest of the thyroid epithelium, the vesicles in the immediate vicinity of round-cell infiltration were almost always of the subtoxic type.

The study of a miscellaneous series of thyroidectomy specimens (second series) showed that the subtoxic variety of vesicle, with or without round-cell infiltration, commonly occurred in the rim of thyroid tissue around adenomata and beneath the capsule in non-adenomatous thyroids. These are exactly the same positions in which round-cell infiltration was first seen. The fact that the subtoxic type of vesicle occurs in the absence of lymphocytic infiltration, while round-cell infiltration alone is very rare, suggests that the development of subtoxic vesicles precedes lymphocytic infiltration.

The functional state of these vesicles is not completely understood. According to Rienhoff (1925) they are not functioning to so great an extent

as the larger vesicles and he suggests that they are resting vesicles with temporarily reduced blood-supply.

Aetiology of Lymphadenoid Goitre.—A considerable number of theories have been put forward :—

1. Inflammatory origin : Against this view are the facts that lymphadenoid goitre only very rarely follows focal sepsis (in contrast to Riedel's thyroiditis, which often follows acute tonsillitis, dental sepsis, etc.) and, secondly, that no organisms have been isolated from any operation specimen.

2. That it follows prolonged iodine medication. A history of this was obtained in only 15 of our cases, so that it is an unlikely factor in the pathogenesis of this condition.

3. A degenerative change. This view has been put by Chute, Eckerson, and Warren (1935) and by Schilling (1945). Vaux (1938) thinks that the condition may result from excessive involution following mild thyrotoxicosis. Joll (1939) rejects this view on the ground that the lymphoid tissue possesses enormous intrinsic activity and does not merely displace something which has undergone shrinkage.

4. Vitamin deficiency. McCarrison (1929) fed rats on a vitamin-deficient diet and produced changes similar to those of Hashimoto's disease. However, no evidence has accumulated to show that there is any vitamin deficiency in humans with lymphadenoid goitre.

5. Eden and Trotter (1942) suggest that lymphadenoid goitre is an exaggeration of the lymphoid infiltration that may occur in toxic goitre.

6. Palmley and Hellwig (1946) believe that Hashimoto's disease is due to overstimulation of the thyroid gland by thyrotropic hormone. The gland fails eventually to produce colloid and round-cell infiltration and atrophy results.

The above theories are not all mutually exclusive. Thus Vaux's view that the condition results from excessive involution following mild thyrotoxicosis is substantially similar to the view, held by Palmley and Hellwig, that it is due to overstimulation by thyrotropic hormone with failure of the gland to respond successfully. Both theories are compatible with the view of Eden and Trotter that the condition is an exaggeration of the round-cell infiltration seen in toxic goitre. All three views postulate a preliminary phase of thyrotoxicosis followed by a stage of relative failure of the activity of the gland with associated round-cell infiltration.

That a thyrotoxic phase is at one time or other a feature of lymphadenoid goitre is mentioned by Polowe (1934), Lee (1935), Chute and others (1935), Poer, Davidson, and Bishop (1936), Lee and McGrath (1937), Vaux (1938), Joll (1939), Eden and Trotter (1942), McSwain and Moore (1943), and Palmley and Hellwig (1946). A few authors failed to find toxic symptoms or history—Hashimoto (1912), Graham and McCullogh (1931), and Schilling (1945).

In the latter two papers, however, their authors remind us that it remains in question whether the "degenerative process" was not preceded by an initial hyperplasia and toxic reaction, which either passed unnoticed by the patient or was forgotten, having occurred so long ago.

The bulk of the evidence, including the material of this study, indicates that a toxic phase is sufficiently frequent to count as an important factor in pathogenesis.

A study of the second series of cases has shown that focal changes histologically indistinguishable from those seen in the various stages of lymphadenoid goitre can very frequently be found in the rim of thyroid tissue immediately surrounding an area of adenomatous hyperplasia, and also immediately beneath the fibrous tissue capsule of the gland. Both locations are sites where compression of thyroid acini would be liable to occur as the result of increase in size of part or whole of the gland. It is reasonable to conclude that the changes are secondary to local ischæmia, and it is suggested that the widespread changes occurring in lymphadenoid goitre are similarly due to ischæmia, probably a relative ischæmia due to the blood-supply being insufficient to meet the increased demands created by the stimulation of the thyroid epithelium by the thyrotropic hormone. The fact that the changes are never seen inside an area of adenomatous hyperplasia, which for its growth and maintenance depends on an adequate blood-supply, is further support for the ischæmic causation of the lymphadenoid changes.

Although degenerative changes in the main thyroid artery branches were not infrequently seen in the cases of the first series, yet no close relation was found between the vascular changes and the degrees of lymphadenoid change, and the incidence of vascular disease was no greater than in the second series of cases where lymphadenoid infiltration was minimal.

Thus there is no definite evidence that an absolute ischæmia due to vascular degeneration is an important factor in the aetiology of lymphadenoid goitre. Nevertheless, the fact that lymphadenoid goitre affects subjects in the later age groups, *see Table III*, when the thyroid vessels are less likely to be able to adapt themselves as readily as those of younger subjects to the increased requirements of a hyperactive gland, is compatible with the view that the changes of lymphadenoid goitre are the expression of relative ischæmia.

Further support for this view is given by the well-known fact that lymphadenoid goitres bleed very little at operation, indicating that in the established phase of the condition there is undoubted diminished vascularity.

Rienhoff's suggestion that the subtoxic vesicles, which we have found to be so common in all stages of lymphadenoid goitre, are vesicles with a reduced blood-supply, corresponds with the above interpretation of the sequence of events leading to lymphadenoid change.

The arguments to support the view that lymphadenoid goitre develops as the result of relative ischæmia may be summarized as follows :—

First, the condition develops in older subjects whose vessels may be presumed to be less capable of responding satisfactorily to a demand for an increased blood-supply.

Secondly, there is in most cases a history of a previous toxic phase, showing that there has in fact been a demand for increased blood-supply.

Thirdly, histological changes similar to those seen in lymphadenoid goitre commonly occur focally in those sites where local ischæmia is most probably present, i.e., around adenomata and immediately beneath the fibrous capsule of the gland.

Fourthly, the changes of lymphadenoid goitre are extremely rarely seen within areas of adenomatous hyperplasia, and such areas may reasonably be regarded as areas of adequate blood-supply since active cellular hyperplasia is being maintained.

Fifthly, diminished vascularity in lymphadenoid goitre is indicated by the fact that such goitres bleed very little at operation.

In conclusion, the examination of the first series of cases has shown that no sharp dividing line can be drawn on histological grounds between toxic goitre with lymphoid infiltration on the one hand and established lymphadenoid goitre on the other; the differences appear to be essentially ones of degree. Examination of the second series has established that sites where local ischæmia may be presumed to have been present frequently show focal changes similar to those in lymphadenoid goitre. Examination of the blood-vessels in both series showed that while degenerative vascular changes were not infrequent in the age groups in which lymphadenoid goitre is common, yet there was no close association between the vascular changes and the presence or absence of lymphadenoid changes. The suggestion is made that lymphadenoid changes are due to a relative ischæmia in a gland subjected to increased thyrotropic stimulation.

SUMMARY

1. The material of the first series of cases comprised 121 cases of thyroidectomy where more than slight lymphocytic infiltration had been found in histological section.

The second series comprised 64 cases in which lymphocytic infiltration was slight or absent.

2. Ten cases of the first series showed the classical picture of Hashimoto's disease. In the remainder round-cell infiltration was less advanced, but the cellular infiltrations of lymphadenoid goitre were present in varying degree.

3. The second series of cases provided evidence that local ischæmia could produce focal development of the histological changes found in the first series of cases.

4. The aetiology of lymphadenoid goitre is discussed. It is suggested that lymphadenoid changes are due to the development of relative ischæmia in a gland subjected to increased thyrotropic stimulation.

We wish to express our gratitude to Professor J. Morley for his encouragement and for the loan of his material; and to Professor S. L. Baker for helpful advice and criticism.

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GAS CYSTS OF THE INTESTINE

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ENTERIC pneumatosis, or gas cysts of the intestine, is a rare condition, the exact aetiology of which is still undetermined.

Characteristically, there are multiple gas-containing cysts in the subserous coat of the intestine; usually the ileum is involved, but not uncommonly the cæcum or ascending colon or, as in the case reported here, the pelvic colon. They vary in size from microscopic to a pigeon's egg and they have, as a rule, a lining of endothelial cells with an outer wall of connective tissue; occasionally the wall is thickened and infiltrated by lymphocytes and foreign-body giant cells. Although they are usually subserosal, they may occupy any layer of the intestinal wall, and in the few cases which have been reported in infants, the cysts have been either mucosal or submucosal. Most of the cysts contain nothing but gas and this gas is said to consist mainly of nitrogen, with small quantities of carbon dioxide or other gases.

The first case was described by Bang in 1876, and it was at first believed (Finney, 1908) that the cysts constituted a distinct type of tumour, the cells of which have the property of secreting gas. Masson affirms that the cysts are dilated lymph spaces, and that the gas is derived from chyle by chemical interaction with acid products of intestinal fermentation, while according to Naeslund and Berglund, the cause is a coli-like bacterium which penetrates the intestinal wall and there assumes gas-forming properties. None of these theories has gained wide acceptance and it is at present generally believed that the condition is best explained by the mechanical theory, i.e., an expulsion of gas from the lumen of the alimentary canal through ulceration of the mucosa. Most of the reported cases have been discovered accidentally during exploration of the abdomen for gastric or duodenal ulcers; in the case reported here, there is strong evidence for the belief that the gas escaped through a simple ulcer of the pelvic colon. There was at the time, in addition, an acute volvulus of this part of the bowel which no doubt contributed to the increased intraluminary tension.

CASE REPORT

J.J., male, aged 63 years, a previously healthy person, was first admitted to the Royal Victoria Infirmary on July 29, 1940, with a two-day history of acute intestinal obstruction. A pre-operative diagnosis of volvulus of the pelvic colon was made and this was confirmed at operation (Mr. S. C. Raw). A simple reduction of the volvulus was performed and there is no mention in the operation notes of the presence of any gas cysts at that time. After this intervention he remained perfectly well until December, 1942. On Dec. 8 he had a severe attack of diarrhœa which persisted for three or four days and was associated with mild colicky lower abdominal pain. Both the pain and the diarrhœa then cleared up and he was all right again until Dec. 17, when the pain recurred, but much more severely, and was associated this time with almost complete constipation. The following day his abdomen began to swell, and this swelling gradually increased until at the time of his admission, on Dec. 21, it had reached truly enormous proportions. There was no vomiting throughout this period.

PHYSICAL EXAMINATION showed a rather poorly nourished individual with a grossly distended and tympanitic abdomen. The distension was so great as to cause a considerable amount of respiratory embarrassment; there was no abdominal tenderness or rigidity. The rectum was ballooned, but otherwise normal. His general condition was fair; T. 98·4°, P. 80, B.P. 110/75.

It was decided that he was suffering from a recurrent volvulus of the pelvic colon; the possibility of a pneumoperitoneum being partially responsible for the abdominal distension was not appreciated and unfortunately a pre-operative X-ray examination was not carried out.

LAPAROTOMY was performed four hours after admission; immediately on opening the peritoneal cavity, there was a loud hiss of escaping gas, and the abdominal wall collapsed like a punctured balloon. The gas did not smell and there was no free fluid. The pelvic colon was grossly dilated, due to a volvulus through 360°. The whole surface of the bowel and its mesentery for a length of about 20 in. was covered and almost obliterated by enormous numbers of gas-filled cysts, varying in size from pin-head to about 2 in. in diameter. Gas could be expressed from one cyst to another. The rest of the bowel was normal. The mesentery of the affected loop was fibrosed and indurated and this scarring had caused the pelvic loop to become U-shaped. A Mikulicz resection of the pelvic colon was performed.

The patient's subsequent progress was uneventful. He was seen again in June, 1947; he remains in good health, has normal bowel action, and has had no further symptoms of any kind.

PATHOLOGICAL REPORT.—The specimen is a portion of the pelvic colon 37 cm. long in the form of a U. The two limbs are joined by thickened mesocolon. The serosa covering it is finely granular and rather pearly. The whole specimen floats in water, due to the fact that all the appendices epiploicæ are distended with gas which is odourless and non-inflammable. The structures mentioned and the whole subserosa, except that over the longitudinal muscle bands, are on section honeycombed by the presence of gas, mainly in the form of bubbles less than 1 cm. in diameter, but a few are up to 3·5 cm. The nature of the gas is uncertain, but it is non-inflammable and is presumably not hydrogen. The bowel contains fæces at one end for a distance of about 6 cm. The rest of the bowel is empty except for some mucus. There are no polypi or diverticula. On the mesenteric border, exactly at the bottom of the U, is a transverse, very narrow, elliptical ulcer 1·2 cm. long. It extends only into the submucosa and the edges are firmly attached to the floor, except at one point, where a probe can be passed readily for a length of 3·0 cm. into the underlying tissues. The cause of the ulcer is not apparent from the gross appearances, but it is clearly not a neoplasm. It is, however, almost certainly the site at which gas from the lumen of the bowel has escaped into the surrounding tissues.

Microscopically, the mucosal lesion is a simple chronic ulcer. Its floor is formed of granulation tissue, and its chronic nature is indicated by the replacement of the muscle by scar tissue. Lying in the granulation tissue are a few minute doubly-refractile foreign bodies, surrounded by foreign-body giant cells. Similar structures are seen not infrequently in ulcerated carcinomas of the

gut and most probably have entered the tissue after the ulcer was formed. There is nothing in the section to indicate the cause of the ulceration. It is evident that the sinus leading from the ulcer could act as a flap valve, permitting gas to pass into the tissues.

DISCUSSION

This case presents many unusual features—an acute volvulus of the pelvic colon, with a coexisting simple ulcer of the affected loop of the bowel; associated multiple gas cysts of the bowel and mesentery; and a large pneumoperitoneum. Self-limitation of the gas cysts with fibrous tissue healing is the usual sequence, but this could hardly have occurred in this case; moreover it was evident that the enormous pneumoperitoneum was at least in part responsible for the distress associated with the abdominal distension. It is interesting to note that in a case reported by Pybus in 1934, X-ray examination showed air between the liver and diaphragm, although there was no perforation. It also showed the outlines of some of the cysts. It is probable that in every case of enteric pneumatosis, some degree of pneumoperitoneum is liable to occur, and if this is excessive, it can give rise to definite symptoms. The statement that "enteric pneumatosis gives rise to no symptoms and is of no clinical importance" (Illingworth and Dick, 1947) is therefore not strictly accurate.

I should like to thank Professor J. H. Barclay for permission to publish this case and Dr. J. G. Thomson for the pathological report.

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A CASE OF INTUSSUSCEPTION CAUSED BY ENDOMETRIOSIS OF ILEUM

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THE case recorded below, in which acute enteric intussusception has occurred in association with endometriosis of the ileum, is of interest from several aspects.

While endometriosis is said by Jenkinson and Brown (1943) to occur in approximately 15 per cent of all women during their active menstrual life, the condition is usually confined to the pelvic organs and rarely affects the small intestine. Masson (1935), reviewing 576 cases of endometriosis which were seen at the Mayo Clinic, found the small intestine affected on only 2 occasions, each time in the ileum. Sampson (1924), in a series of 67 cases, found the ileum affected once. Jenkinson and Brown (1943), who reported on 117 cases, and Keene and Kimborough (1930) in their 118 cases, did not find the small intestine affected in any.

CASE REPORT

Mrs. W., aged 37, the mother of two children, was seen by one of us (K. C.) in consultation on Oct. 22, 1946. She complained of increasing dysmenorrhœa since the birth of her last child eighteen months previously. In the last few months she had had three or four attacks of colicky abdominal pain, three of these attacks corresponding with a menstrual period, the final attack occurring two weeks after a period. The pain was intermittent, severe, accompanied by vomiting and dry retching, but flatus was passed and at no time were the bowels constipated. The attacks lasted six to eight hours and necessitated the administration of morphine ¼ gr. twice to effect relief. In the third attack she was seen by a gynæcologist, who noticed a mass in the abdomen. An enema was administered and the mass disappeared.

On Oct. 22, two days after her last attack, she was examined. Her abdomen was flaccid and she had slight tenderness in both iliac fossæ but no muscle guarding. On vaginal examination the uterus was found to be anterior and small, and in the left fornix a mass about 6 cm. in diameter was palpable and tender. The diagnosis made was a left ovarian cyst which was undergoing torsion at times and causing the extreme pain.

On Oct. 29 she had another attack of pain commencing at 11.30 a.m. She was admitted to hospital and seen at 5.30 p.m. She was extremely shocked, sweating, and vomiting. Her temperature was subnormal and her pulse 104. She was propped up in bed with her knees elevated and any attempt to lower her head caused extreme pain. The abdomen was flaccid and almost concave in appearance and no mass was palpable. Per rectum a soft fluctuant mass about the size of a grape-fruit was palpable in the pouch of Douglas and was very tender. Torsion of the pedicle of an ovarian cyst was diagnosed and laparotomy performed at 7.30 p.m. Intravenous saline was commenced and the patient placed in Trendelenburg position. Immediately this was done the mass appeared on the right side of the abdomen.

On opening the abdomen through a median subumbilical incision the mass was found to be an ileo-ileal intussusception into the cæcum. The mass was irreducible and the caput cæci was œdematous and cyanotic in appearance. In the pelvis there were bilateral ovarian cysts—the left larger than the right—which both gave the naked-eye appearance of endometrial cysts.

The affected portion of the ileum was resected together with 6 in. of proximal ileum and a portion of the cæcum on account of its doubtful viability. A side-to-side anastomosis of the ileum to the ascending colon was performed and the abdomen closed. One litre of blood was transfused and the patient made an uninterrupted recovery. The bowels opened without assistance on the fourth day and the patient is now well except for the fact that she has about three bowel actions a day. The reason for this has not yet been ascertained.

The portion removed was examined after fixation in 10 per cent formalin. The distance from the neck of the

intussusception to the apex was approximately 25 cm. The proximal two-thirds of the mass was curved so that the mesenteric attachment to the small intestine was on the concavity. The distal third of the intussusceptum, which lay in the cæcum, was curved in the opposite direction. About 8 cm. from the neck a nodule was found in the wall of that part of the ileum which formed the entering layer of the intussusceptum. It was on the side opposite the mesentery. The nodule was 1 cm. in diameter and of pale yellow tissue with some fine white strands and two or three small red spots. It appeared to be directly in contact with the mucous membrane on one side and the peritoneum on the other.

FIG. 79.—Wall of small intestine showing endometrial tissue in the muscle coat. (× 16.)

Examination of a microscopical preparation of the mass shows that glandular tissue is present in the muscle coat of the small intestine (*Fig.* 79). In some parts, one of which is indicated by the arrow, this tissue is arranged in a compact mass. In other parts the glands are more widely separated and some of these are of comparatively large size.

Under higher magnification (*Fig.* 80) the glands and supporting connective tissue closely resemble normal endometrium. The new tissue is surrounded by involuntary muscle of the small intestine. The microscopical picture is one of endometriosis.

DISCUSSION

The enteric type of intussusception has more frequently an obvious anatomical cause than any other (Perrin and Lindsay, 1921). In the case reported above there can be little doubt that the endometriosis initiated the invagination. Usually the mass which is the cause is situated at or near the apex of the intussusception; however, it may be found near the neck (Wardill, 1925), as in this instance. No report of intussusception caused by endometriosis has been found in the available literature.

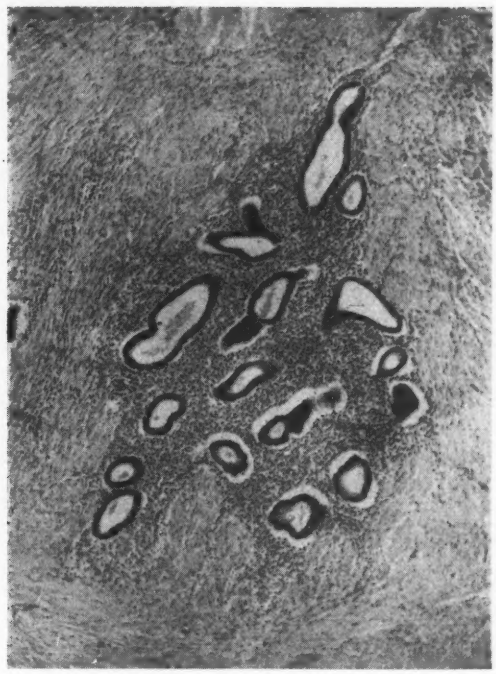

FIG. 80.—The part of *Fig.* 79 indicated by the arrow is here shown under higher magnification. (× 40.)
(This is not taken from the same section as in *Fig.* 79, but from another section in the same series.)

SUMMARY

1. A case is reported in which acute enteric intussusception has been caused by endometriosis of the ileum.

2. The relative infrequency with which endometriosis affects the ileum is noted.

We wish to thank Mr. S. Woodward-Smith for the microphotographs.

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CONGENITAL OBSTRUCTION OF THE COLON IN AN ADULT

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CONGENITAL obstruction of the intestinal tract is a subject which has been exhaustively discussed in the surgical literature; as it affects the colon, however, the condition has been neglected in proportion to its rarity.

The case described in this paper is of particular interest, both in so far as the anomaly involves the colon and also in that it was discovered in an adult.

CASE REPORT

HISTORY.—E. H. L., a male aged 41, was admitted to Dunedin Hospital under the care of Mr. James A. Jenkins.

The patient complained of lower abdominal pain which had troubled him since the age of 14. His childhood had been healthy. He was admitted to this hospital at the age of 27 (Feb. 11, 1933), and submitted to operation for acute appendicitis: the pathological report on the excised organ described "chronic appendicitis with recent acute exacerbation".

Following this operation the patient remained well for twelve months, but over the subsequent four years he experienced occasional attacks of colicky pain and distension, relieved by the passage of flatus. By 1938 these attacks had become so frequent and so severe that the patient was re-admitted (Jan. 19, 1938) to hospital with the provisional diagnosis of intestinal obstruction. An acute attack of colic lasting for two and a half hours, and described by the observer as "agonizing", was noted on this occasion and a "rounded smooth mass" was felt in the right iliac fossa. A plain radiograph of the abdomen was reported negative except for spina bifida of the fifth lumbar vertebra, with unilateral sacralization on the left side.

Laparotomy was performed, and no abdominal pathology was detected apart from "a large lax cæcum", which was plicated. This operation took the form of a series of seromuscular sutures introduced transversely into the wall of the cæcum so as to reduce its calibre. The diagnosis arrived at was one of "cæcal stasis".

Subsequently the patient remained well for a few months: symptoms then recurred in exactly the same form as before, and between February, 1939, and August, 1946, the patient was admitted to various departments of the hospital on fourteen occasions. Each time the diagnosis made was one of "functional abdominal pain" or "neurasthenia". It would appear that in spite of the patient's pathetic insistence that his were not imaginary ills, he did in fact present a progressively more neurotic disposition, and was on one occasion diagnosed by a psychiatrist as "advanced incurable neurasthenia". The basic symptomatology, however, remained constant, and was reiterated each time the patient presented himself.

CLINICAL FEATURES.—The patient was first seen by the writer on Aug. 29, 1946. He described his *pain* as being intermittent, irregular, and unrelated to food. Its location was across the lower abdomen, and in the R.I.F. It was colicky in nature and sometimes so severe as to double him up. Attacks of pain usually lasted a few hours: longer attacks were associated with *constipation* lasting from four to seven days. Relief, when it came, was usually due to enemata, and coincided with the passage of large quantities of *flatus*. *Vomiting* was exceptional, but *dyspepsia* was a constant complaint, meals being followed by pain, flatulence, and heartburn. *Melæna* appears to have occurred on several occasions, and *borborygmi* troubled the patient incessantly.

Distension was never observed in generalized form, but both the patient and successive observers described a gurgling sausage-shaped swelling in the R.I.F. The records show that this swelling had been observed in hospital both before and after the operation for plication of the cæcum. Latterly the patient had learned to massage the swelling and so cause its disappearance, which always coincided with the release of flatus and cessation of the pain.

PHYSICAL EXAMINATION.—The patient is a lean, sallow individual, worried about his condition, but not obviously neurotic. Routine examination is negative except for the demonstration of a swelling in the R.I.F. This varies from day to day: when present it is found to be smooth and rounded in shape, mobile, tense in consistence, resonant to percussion, and noisy on auscultation.

RADIOLOGICAL EXAMINATION.—A barium meal was reported on by Dr. Charles Begg: "The stomach, duodenum, and small intestine are normal. About the junction of the cæcum with the ascending colon there is a stricture where the lumen of the gut is reduced to the size of a lead pencil. Gas and barium could be squirted through this constriction by pressure on the cæcum. The barium passed through in normal time limits".

OPERATION.—Laparotomy was undertaken by Mr. James A. Jenkins on Sept. 13 under spinal and local anæsthesia. A right oblique muscle-cutting incision was made, excising the old appendicectomy scar. Exploration of the cæcum and ascending colon revealed no external evidence of stricture, but high up on the ascending colon there was marked thickening and some induration suggestive of chronic inflammatory change. Enlarged glands were found at the ileocæcal junction, and also below and medial to the hepatic flexure. Excision of the right colon was performed and a lateral anastomosis made between ileum and transverse colon.

POST-OPERATIVE PROGRESS.—The patient made an uneventful recovery and was discharged on Oct. 2. He was re-admitted three months later, complaining once more of abdominal pain. This had apparently recurred three weeks after his discharge, gradually increasing in frequency and severity until he had three or four attacks every day. The pain was of a colicky umbilical type and was associated with a swelling near the upper end of the wound. No vomiting, but bowel function was irregular.

On Feb. 7, 1947, the abdomen was re-opened and the blind stump of ileum in the vicinity of the anastomosis was found to be grossly distended. This was excised and the bowel closed. The anastomosis was patent, and the suture line intact. Convalescence was uncomplicated, and the patient discharged fifteen days later.

When re-examined on May 29, the patient is found to be free from pain, and to have regular bowel function. His outlook is cheerful, and he is well satisfied with the result of the operation.

PATHOLOGICAL REPORT.—"The specimen (*Figs.* 81-83) consists of terminal ileum, cæcum, ascending colon, and part of transverse colon. The appendix has been removed at some previous date. The cæcum is much shrunken and fibrosed. There is a transversely placed diaphragm across the ascending colon 2 in. above the ileocæcal junction. This practically occludes the lumen, having only a small central perforation about ¼ in. in diameter. Four to five inches above the ileocæcal valve the wall of the colon is grossly thickened, and just proximal to this thickening is another diaphragm which almost

occludes the lumen. Sections from the area of thickening show the mucosa to be for the most part normal. There is a considerable increase in the fibrous tissue of the submucosa, and at one point there is a small acute abscess, the mucosa at this point being ulcerated. This is a non-specific inflammation associated with two congenital diaphragms of the ascending colon." (Professor D'Ath.)

DISCUSSION

The lesion described was undoubtedly the cause of the symptoms from which the patient suffered. The long history of intermittent intestinal obstruction

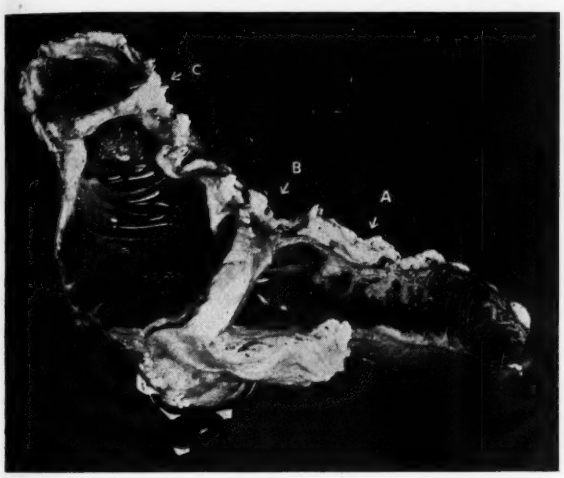
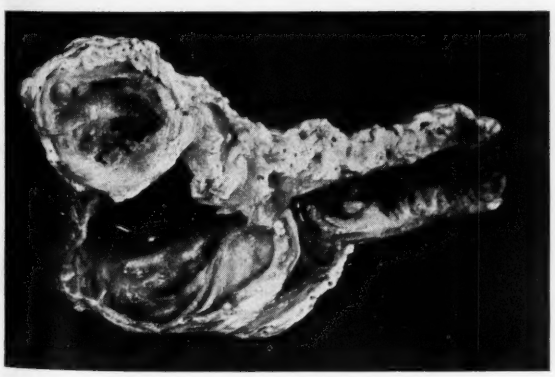


FIG. 81.—Terminal ileum, cæcum, and ascending colon, showing: A, Ileocæcal junction; B, Lower diaphragm; C, Upper diaphragm.

with distension; relief with subsidence of the swelling and the passage of flatus; the radiologic findings; and the patient's relief following operation, leave



FIGS. 82, 83.—Oblique views showing the diaphragms. It will be noted that the cæcum is shrunken and fibrosed and that the bowel between the two diaphragms is slightly distended.

little doubt on this score. Indeed, it is surprising that the patient could have led a comparatively normal life for so long while harbouring such a lesion.

The following points appear to merit further discussion:—

1. **The Nature of the Lesion.**—It is considered important to stress the congenital nature of the diaphragms. The only other factors that could conceivably be responsible are (*a*) Inflammatory; (*b*) Operative.

The patient had no history, nor was there pathological evidence, of inflammatory lesions of the bowel other than appendicitis, which could not have caused one, still less two, of these diaphragms. There is indeed thickening of a chronic inflammatory, but non-specific, nature in the region of the upper diaphragm. Such changes, however, are absent round the lower diaphragm, which consists solely of a thin layer of fibrous tissue covered on each aspect with healthy mucosa, and which unites around its periphery with normal bowel wall. The inflammatory element described must therefore be regarded as secondary to chronic obstruction and in no way causative.

The patient had been submitted to two abdominal operations prior to excision of the right colon. The first, a straight-forward appendicectomy, may be dismissed: the second, plication of the cæcum, may be regarded with more suspicion.

However, as (i) the identical symptom complex was present before and after plication; (ii) a distended cæcum (or ascending colon) was observed before, during, and after this operation, and indeed constituted the only indication for its performance; (iii) one vertical plication of the cæcum would hardly cause two transversely placed diaphragms of the ascending colon; and, finally, (iv) the diaphragms are thin membranes with only very small central perforations—these points rule out any possibility that operative interference could have been an aetiological factor.

The congenital nature of the diaphragms cannot therefore be held in doubt.

2. **The Rarity of the Lesion.**—The incidence of congenital obstruction of the intestine, excluding anal imperforation, is estimated at 1 in 20,000 births (Farr and Brunkow, 1925). The cases reported in the literature as involving the colon, and comprising 10 per cent of the total, include the following conditions:—

a. Microcolon.—The whole colon, or rarely a segment of it, is of extremely diminutive calibre, but in true microcolon a lumen is present throughout. The condition may be found independent of obstruction at higher levels, or, much more frequently, it is secondary to atresia of the small intestine (Greig, 1925). This group comprises the majority of cases reported as congenital obstruction of the colon.

b. Atresia.—There is complete absence of a segment of bowel, its representation by a cord of tissue devoid of a lumen, or its occlusion by a complete membranous diaphragm. Some cases of so-called microcolon fall within this group, when the whole colon is reduced to a solid cord of tissue.

c. Stenosis.—There is incomplete obstruction, taking the form of a stricture of the bowel wall, or of a perforated diaphragm spanning the lumen.

d. Complete Absence of the Colon.—Considering the relatively greater incidence of microcolon, it is unsatisfactory that all these conditions should have been lumped together in the early papers describing large series of cases of congenital bowel obstruction. Thus Davis and Poynter (1922) collected 392 cases, of which 39 were described as affecting the colon, no further classification being given.

Dissatisfied with the position, Heckel and Apfelbach (1927) collected from the literature a series of cases which excluded microcolon. They were able to report 29 examples of atresia, and added 1 case of their own. In all, the lumen of the bowel was completely interrupted: in 24 by absence of a segment of bowel, and in 5 by a complete membranous diaphragm.

Since 1926 there have been reported, so far as I can discover, 21 further examples of atresia or stenosis of congenital origin. The original papers available to me are tabulated in *Table I*, and show,

Table I

Year	Author	No. of Cases of Congenital Obstruction	No. of Colon Cases	
			Atresia	Stenosis
1926	Sheldon	34	3	2
1929	Baty	12	1	
1931	Webb and Wangensteen	17	1	
1930	Popper		1	
1937	Bowers and Cook		1	
1941	Ladd and Gross	74	2	1
1944	Richdorf			1
			9	4

in a total of 13 cases, 9 examples of atresia and 4 of stenosis. It is thus apparent that stenosis of the colon is one of the least common forms of congenital intestinal obstruction.

3. The Rarity of Double Stenosis.—The case presented in this paper shows two well-developed diaphragms, each of which almost completely occludes the lumen of the bowel. I have not been able to discover in the literature reference to a similar occurrence, while Ladd and Gross state (1941) " In our experience, no case has had more than one stenosis, which is in contrast to the findings in atresia ".

4. The Age of the Patient.—Congenital occlusion of any part of the gastro-intestinal tract is an extremely rare cause of symptoms in adult life. Instances of such an event, however, are recorded by Carling (1910), Spriggs (1912), Cannon and Halpern (1922), Judd and White (1929), and Ladd and Gross (1941), the site of obstruction being in each case the small intestine, and the age of the patients ranging from 9 to 47.

In papers which are not available here, the following cases involving the large bowel in childhood or adult life have apparently been reported: Duroux (1930) described microcolon in an adult; Caldas (1933), a case of " extensive atresia " in an adult; and Silva (1941), congenital stricture of the colon in a child.

5. Causation.—The commonly accepted aetiology of both atresia and stenosis concerns the epithelial proliferation which takes place in the second month of fœtal life, to the extent of obliteration of the lumen, which later becomes re-established by absorption of excess tissue. Incomplete reabsorption can readily be understood to leave behind such diaphragms as are described in the present case.

SUMMARY

A case is described in which chronic intestinal obstruction in an adult was found to be caused by two congenital diaphragms of the ascending colon.

I am indebted to Dr. James Thompson, Medical Superintendent of Dunedin Hospital, and to Mr. James A. Jenkins, for permission to publish these notes.

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CÆCAL FÆCOLITH

A REPORT ON TWO CASES

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THOUGH every student is familiar with the small fæcal concretions found in the appendix, large fæcoliths and other forms of fæcal accumulation are rarer and have attracted little attention in this country, most recent references in the literature coming from warmer latitudes, notably France, Italy, and South America.

The earliest reference was made by Guillenceau, who, in 1620, described a case of dystocia due to a rectal fæcolith. In 1813 Foumier described a recto-vaginal fistula caused by the discharge per vaginam of a rectal fæcolith in a case with imperforate anus. In 1816 Bright recorded a large tumour in the left iliac fossa cured by calomel, while Duncan, in 1819, described a case of " cancer of the bowel " cured by a purge.

These classical cases all illustrate the cause of these masses, which is essentially bowel stasis, giving rise to accumulation of fæcal matter which is subjected to a process of desiccation. If the accumulation is relatively rapid, and fluid absorption slow, the mass remains large and pultaceous, being referred to as fæcal accumulation or impaction, or as a fæcaloma, stercoroma, or coproma. Digitally it can be distinguished from a neoplasm in that, like putty, it can be indented (Hofmokl, 1896). If accumulation is slower and desiccation faster, the mass may become very hard and dry—the so-called fæcolith. It is not generally recognized that these stones, like others elsewhere in the body, even if not primarily formed of radio-opaque material, may become so from the precipitation of mineral salts from the medium by which they are surrounded.

Both the hard and soft varieties are commonest in the rectum or sigmoid and are usually due to habitual constipation, but may possibly be secondary to bowel strictures, congenital, inflammatory, malignant, or functional, as in Hirschsprung's disease.

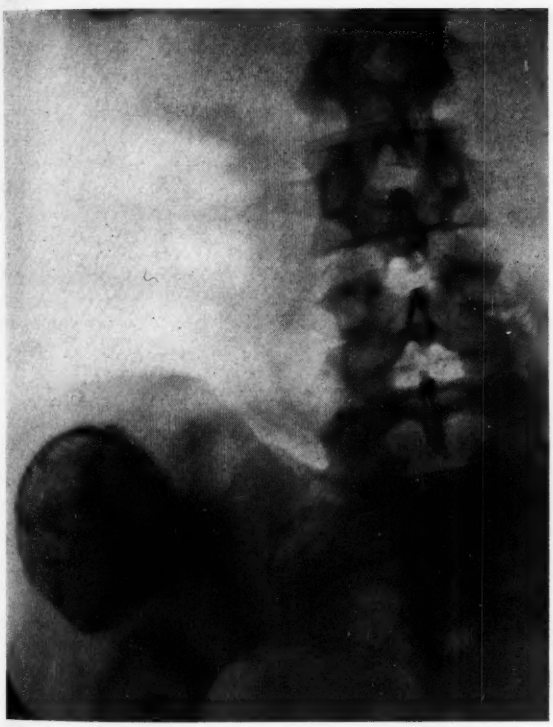

FIG. 84.—*Case* 1. Abdomen, plain film.

FIG. 85.—*Case* 1. Barium enema showing fæcolith filling the cæcum.

Rosenrauch (1933), in his review of the subject, considers fæcoliths and fæcalomas elsewhere than the rectosigmoid region to be of the greatest rarity. It is for this reason that the two cases to be described, together with their radiological features, seem worthy of record.

CASE REPORTS

Case 1.—Mrs. E. P., aged 53, housewife, was admitted on Dec. 31, 1946, having been well until eight months before, when attacks of colicky mid-abdominal pain with audible borborygmi began. These attacks occurred almost daily thereafter, with anorexia, nausea, and on two occasions, vomiting, but no alteration of the bowel habit.

Examination was normal apart from the abdomen, which was moderately distended. In the right iliac

fossa, a spherical mass 1½ in. in diameter was felt, stony hard, and mobile, being displaceable upwards almost to the costal margin and downwards to the pelvis. There was neither tenderness nor rigidity. A plain radiograph of the abdomen (*Fig.* 84) showed an almost spherical shadow in the right iliac fossa corresponding to the palpable lump. A barium enema (*Fig.* 85) showed barium surrounding the mass, which was apparently in the cæcum, and which was considered most probably to be a fæcolith. It was further supposed that both the fæcolith and the subsequent obstructive symptoms were due to an occlusive lesion of the ascending colon.

The symptoms of obstruction increased, with vomiting necessitating the passage of a Miller-Abbott tube.

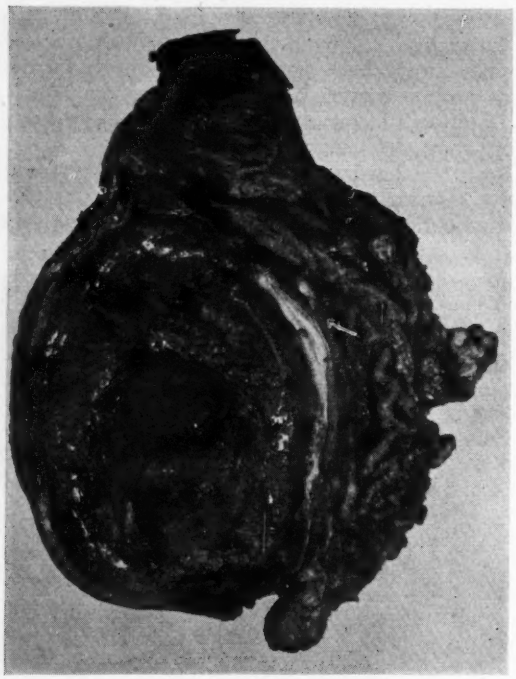

FIG. 86.—*Case* 1. The specimen.

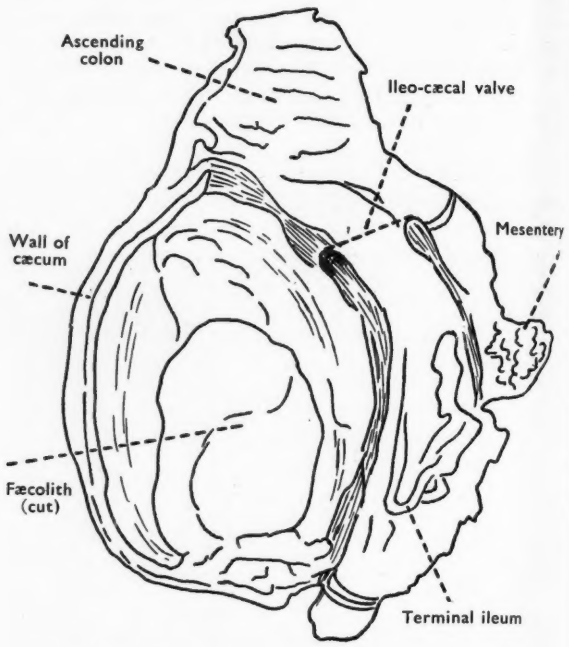

FIG. 87.—*Case* 1. Diagram of specimen.

FIG. 88.—*Case* 2. Abdomen, plain film.

FIG. 89.—*Case* 2. Barium enema.

On Jan. 1, 1947, the abdomen was explored through an oblique muscle-cutting incision in the right iliac fossa, and the hard mass was found to be inside the cæcum, which, with the ascending colon, was mobile on a long mesentery. The collapsed terminal ileum prompted a search for the real obstructing lesion, which turned out to be an area of regional ileitis, 1½ ft. from the ileo-cæcal valve, with enlarged fleshy mesenteric glands. The involved segment of ileum with a wedge of mesentery containing the glands was resected and side-to-side anastomosis performed. Likewise the cæcum was resected and side-to-side anastomosis performed between the terminal ileum and the ascending colon.

From this the patient made an uninterrupted recovery, and was well and symptom-free four weeks after discharge from hospital.

The focus of Crohn's disease is regarded as unrelated to the fæcolith, and will not be described in detail. The cæcum (*Figs.* 86, 87) was examined by Dr. G. Cunningham, and was seen to be distended by the fæcolith, 6 cm. in diameter. The fæcolith filled the cæcum but did not encroach on the ileo-cæcal valve. It had grown to such a size that it had converted the cæcum virtually into a diverticulum from which it could not escape. The cæcal walls were not inflamed. The appendix (not shown on the prepared specimen) has a normal lumen, as can be seen in *Fig.* 85.

The stone itself was light in weight, brown in colour, and cut easily, like papier mâché. On section it was roughly laminated.

Case 2.—Mrs. A. H., aged 60 years, food server, attended the Casualty Department in March, 1947, for treatment of an attack of acute gastro-enteritis. She had had two similar attacks within three months, each, as

FIG. 90.—*Case* 2. Fæcolith.

did this one, subsiding in 2–3 days. She had no other symptoms, her bowels usually acting daily without straining, helped at times by salts.

Examination was normal except in the abdomen, where a stony hard spherical swelling, about 2 in. in diameter, was readily palpable in the right iliac fossa. Great was my surprise, when called to see the case, to find a lump so similar in most respects to the last one and so soon afterwards. It, too, could be pushed up almost to the costal margin and down to the pelvis. It was not tender, and there was no rigidity. Rectal and vaginal examinations were normal.

Remembering the radio-opacity of the previous stone, likened by a medical colleague who saw the case to a "full moon in the pelvis", a plain film was ordered (*Fig.* 88) which was reported on as either a calcified fibroid (unlikely from the physical signs), or a cæcal fæcolith. Barium meal, followed through, showed the mass apparently surrounded by terminal ileum, but no definite evidence that it was within the lumen (*Fig.* 89).

She was admitted to Hospital on April 9 and operation undertaken by Mr. E. G. Tuckwell on April 11, 1947. The right iliac fossa was explored through a muscle-splitting incision and the cæcum delivered. It had a long mesentery and was seen to contain the mass within its lumen, almost exactly as in the previous case. After routine appendicectomy, cæcotomy was performed and a hard, relatively light fæcolith of dark brown colour was removed (*Fig.* 90).

Barium enema, performed eight weeks later, showed a normal cæcum, but films taken 48 hours after barium meal showed a considerable residue of barium in the cæcum.

DISCUSSION

Diagnosis.—The diagnosis in left-sided fæcal tumours has often been difficult as they have been mistaken for ovarian or uterine swellings or for carcinoma of the colon. In both the present cases the mass was obviously ileocæcal. The roundness, hardness, and discreteness of the swellings, coupled with their mobility, tended to eliminate carcinoma. On clinical grounds alone the distinction is difficult. H. D. Cronyn (1941) described a fæcolith 3 in. in diameter removed by enterectomy from the right side of the colon of an African which was deemed neoplastic pre-operatively. In this case, no radiograph was taken. Most standard works on radiology do not mention the potential radio-opacity of fæcoliths. Brun and his colleagues (1931), however, described a rectal fæcolith the size of a tangerine visible on a radiograph, while Dresser (1929) described a similar one of grape-fruit size, and Ross Golden (1941) shows a plate with 4 radio-opaque rectal fæcoliths. Kerley (1936) does not mention this, but states that radiologically a fæcolith (radio-translucent) may be distinguished from a carcinoma in that a barium enema, under pressure, may move the stone, which, after evacuation, may remain coated with the medium. He shows a plate of a fæcolith in the ascending colon thus displayed, closely resembling the shadows in *Figs.* 84 and 88. These cases suggest that when a large fæcolith is suspected, X-ray examination, both plain and contrast, may enable the diagnosis to be confirmed.

Aetiology.—The cause of fæcolith formation in these cases is obscure. The cæcum is a cul-de-sac which is normally emptied by taking part in the mass movement of the rest of the right colon. It is probably significant that each of these patients had a long mesocolon which is so often associated with abnormal peristalsis, as for example in intussusception and volvulus. In neither patient was there any evidence of chronic constipation and there was no local cause to delay cæcal emptying.

Cæcal stasis has been described by Shanks and his colleagues (1938) as a rare, symptomless condition detected after a barium meal by the delayed emptying of the cæcum, although the rest of the colon is normal. They quote Barclay's suggestion (1936) that it is due

to a local peristaltic defect—the wave may be incomplete so that the cæcal content, instead of being passed on *en masse*, tends to be partially forced back into the cæcal cul-de-sac. They consider this of little clinical significance—although it seems to me that such cases are those most likely to develop fæcoliths of the cæcum. The follow-through of *Case* 2, three months after operation, did show a considerable hold-up of barium in the cæcum 48 hours after the meal, which then had largely passed the rectum. This, as far as one case is able, seems to support this theory.

Treatment.—Unrelieved, these masses must, by their bulk, eventually cause local catarrhal inflammation, as in Cronyn's case. Spieler (1936) described a similar case which he termed "typhlitis stercoralis". The cause, in this case a fæcaloma, was shifted by castor oil. In the case of the masses here described there seems no doubt that they could not be moved out of the cæcum by purging. Operation must be undertaken, not only to settle any diagnostic doubt, but to remove the mass, and so to prevent future sepsis, which might end in perforation. There seems no danger of occlusion in these cases: the fæcolith is confined to the cæcum and does not encroach in the ileocæcal valve.

If the theory of causation suggested is accepted, resection of the cæcum would seem the logical treatment, as in *Case* 1, so as to remove any chance of recurrence. Enterotomy, as in *Case* 2, would seem to be justified in most cases, however, in view of the low morbidity of these masses and the known mortality of resection.

SUMMARY

Two cases of fæcolith of the cæcum are described, together with their distinctive radiological appearances. There is evidence to suggest that they are caused by localized cæcal stasis. Treatment by enterotomy is preferred.

I wish to express my thanks to Professor J. Paterson Ross, Director of the Surgical Professorial Unit at St. Bartholomew's Hospital, for help and advice and permission to publish these cases, and to the staff of the Photographic Department of St. Bartholomew's Hospital for the illustrations.

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TWO CASES OF VOLVULUS OF THE STOMACH IN A DIAPHRAGMATIC HERNIA

BY H. A. KIDD
SENIOR SURGEON, ST. HELIER HOSPITAL, CARSHALTON

DIAPHRAGMATIC herniæ are not uncommonly discovered as an incidental finding, but few cases have been recorded of strangulation occurring in children.

They may be classified as:—

1. Congenital:
   a. Hiatus pleuroperitonalis (foramen of Bochdalechi);
   b. Dome of diaphragm;
   c. Œsophageal hiatus;
   d. Foramen of Morgagni;
   e. Absence of part of diaphragm.
2. Acquired.
3. Traumatic.

The following cases are examples of the congenital type, the first being through the left hiatus pleuroperitonalis, the second through the para-œsophageal hiatus. Both cases showed the signs of strangulation of the stomach alone, which Rodney Maingot states has not been recorded (*Post-Graduate Surgery*, **3**, 5115), and in both the stomach had rotated on the long axis, the greater curvature having become the highest point in the hernia.

CASE REPORTS

Case 1.—M. P., aged 15 months, was admitted to St. Helier Hospital, Carshalton, on June 2, 1945.

HISTORY.—Normal health until 12 a.m. on day of admission, then had attack of pain, cyanosis, and rapid respirations, but no vomiting.

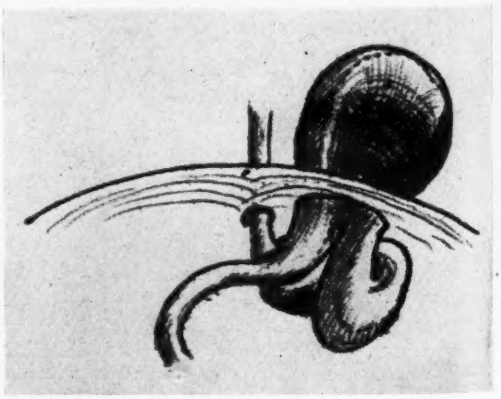

FIG. 91—*Case* 1. Sketch showing left pleuroperitonalis hiatus hernia, containing stomach, with obstruction due to volvulus.

ON EXAMINATION.—

Physical Signs: Diminution of air entry, left base.

Radiography: Left-sided diaphragmatic hernia, with about half of the stomach in the left thorax, causing

compression of the left lung and displacement of the heart to the right.

The diagnosis was made by the pædiatrician, Dr. O'Reilly, and operation performed that night.

AT OPERATION.—Under ethyl chloride and ether (Dr. Goodhart), and local infiltration with 1 per cent novocain, a left transverse upper abdominal incision was made. The spleen presented and the stomach was found to be rotated on its long axis, and so folded that a volvulus was present. (*Fig.* 91.)

One finger could be passed with ease into the thorax, but reduction was not possible, owing to the dilatation of the stomach, until a small opening had been made into the stomach and a catheter passed to aspirate the gas and fluid. The hernia also contained part of the transverse colon and a loop of small intestine: they were not obviously obstructed. The opening in the diaphragm was about 2½ in. long and was closed with a continuous silk suture. There was no peritoneal sac.

Convalescence was uneventful, apart from pyrexia, associated with the collapsed lung. The child was discharged on June 30 with the lung fully expanded. There has been no recurrence.

FIG. 92.—Radiograph, after injection of lipiodol into œsophagus, showing dilated œsophagus and filling of portion of fundus of stomach, subdiaphragmatic in position and not involved in hernia.

Case 2.—Baby C., aged 14 days. Born in the Maternity Department of St. Helier Hospital. Birth weight 7 lb. 9 oz. Regurgitated feeds on third day and vomiting became progressively worse. Eumydrin was given.

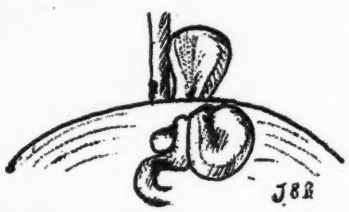

FIG. 95.—Sketch showing para-œsophageal hernia with herniation and volvulus of stomach. Gastric obstruction caused by volvulus, not by obstruction at hernial orifice.

June 16, 1947: 12 noon, X-ray examination, after lipiodol swallow, showed a hold-up at the lower end of the œsophagus. (*Figs.* 92, 93.)

5 p.m., Barium swallow given: radiograph showed that two-thirds of the lipiodol was in the stomach and one-third had passed on into the small intestine. The barium ran easily into the stomach, but would not pass the pylorus. (*Fig.* 94.)

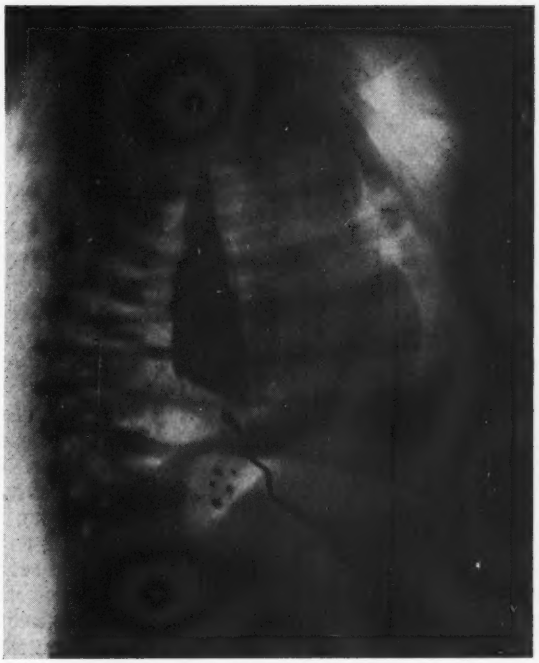

FIG. 93.—Lateral view, showing hold-up of lipiodol in œsophagus and stomach; the hold-up in subdiaphragmatic portion of stomach, proximal to volvulus.

FIG. 94.—One-third of lipiodol in small intestine and barium filling œsophagus and stomach.

June 18: Operation under ethyl chloride and ether (Dr. Goodhart). A left paramedian incision was made. Para-œsophageal herniation of the central portion of the greater curvature of the stomach was found. (*Fig.* 95.) Reduction was achieved after mobilization of the left lobe of the liver. The small hernial orifice was not closed off.

Convalescence was uninterrupted. The baby was discharged on July 5, weight 7 lb. 6 oz., and has remained well.

COMMENTS

1. In both cases the greater curvature became the highest point in the hernia, causing a volvulus effect. This might have occurred through the greater curvature being pushed up by dilatation of the transverse colon, until the diaphragm hiatus was reached; the negative pressure in the thorax may then have sucked the stomach through the diaphragm.

2. Both cases were successfully treated by operation through an abdominal approach.

3. Strangulation of the stomach by volvulus in a diaphragmatic hernia is a rare condition, and it would appear that the obstructive symptoms were due to the rotation of the stomach on its long axis rather than to constriction by the diaphragm at the hiatus.

A STUDY OF PEPTIC ULCER IN NIGERIA

By MAURICE ELLIS

Historical Introduction.—Blair Aitken (1933) was the first to call attention to the presence of peptic ulcer amongst natives of Nigeria. He pointed out that many cases of dyspepsia seen in the Out-Patient Department of the African Hospital, Lagos, Nigeria, previously attributed to helminthiasis, were in fact due to peptic ulceration. His paper was valuable in so far as it showed that peptic ulceration was not the great rarity it was traditionally supposed to be and that many cases could be found, if only a search was made. Since Blair Aitken's paper, no further contribution dealing directly with peptic ulcer in Nigeria has been published. Two further contributions to the study of dyspepsia in Nigeria have, however, appeared.

Up till 1934, no fractional test-meals had been carried out in Nigeria as part of the investigation of dyspeptic patients. I was posted to the African Hospital in Lagos for a few months in 1934. During this time I carried out a large number of test-meals on non-dyspeptic patients. My results were published in the local medical journal—Ellis (1934). Working independently, Rose carried out a similar investigation among the natives of Northern Nigeria and his paper was also published in the same local medical journal—Rose (1935). My results, confirmed by Rose, showed that the gastric response of natives, living on their starchy diet, very different from the European diet, was the same as the gastric response of the European. Bennett and Ryle, in their original investigation, showed that the charts, now used as standard for recording the results of fractional test-meals, indicate the limits of normal secretion for 80 per cent of the Guy's medical students, on whom their tests were carried out. In my series of cases, 80 per cent also showed secretion within these limits. The Bennett and Ryle chart is, therefore, now in standard use in Nigeria.

History of Present Study.—Whilst temporarily stationed in Lagos in 1934, I was able to treat and assist in the treatment of a certain number of cases of peptic ulcer in the African Hospital. On leaving Lagos for a post up-country, I was able to treat a further small number of cases.

On my posting to Lagos in 1937, I then had to diagnose and treat a much larger number of cases of dyspepsia, who were now coming to hospital, usually asking for operation. At the end of a six-year period in August, 1943, a considerable amount of clinical material had accumulated. A general review of this material might form some contribution to the study of peptic ulcer.

In the investigation and diagnosis of dyspepsia cases suspected of peptic ulcer, we are at some disadvantage in Nigeria. In the first place, there is no practising radiologist. A series of pictures taken by a technician, after the patient has swallowed a barium meal, is the best we can achieve. As such pictures are regarded as little, if any, use, radiographs could not be used as an aid to diagnosis.

Secondly, Hurst has stressed the value of occult-blood tests in the diagnosis of active peptic ulcers. As infestations with helminths, particularly minor infestations of hookworm, are so common in Nigeria, the value of this test is doubtful and the test was not carried out as routine.

In diagnosis, therefore, reliance had to be placed on the history and the clinical picture, with such help as the fractional test-meal might occasionally give. In the early cases, the diagnosis could often be made by the patient showing visible gastric peristalsis. In the absence of this, the case history was all that was available. In these early cases, the histories of cases proved to be peptic ulcer showed little in common. One reason for this was, of course, that the history usually had to be taken through interpreters, who had a habit of translating what they thought the patient meant.

Finally, I devised a questionnaire form (a copy of which is given in the appendix) and this questionnaire was filled in for each patient. The physical findings, the operation findings, and the after-history of the patient were filled at the back of the form. This questionnaire was found to give definite answers to many points to allow comparison, without introducing too many leading questions. This questionnaire was used for my last 80 chronic cases.

INCIDENCE

General Incidence of Ulcer.—In a period of six years, ending August, 1943, 191 cases of peptic ulcer have come to operation in the African Hospital,

Lagos. An even greater number of cases have been treated as peptic ulcer in the medical wards by diet and alkalis. But the diagnosis of these medical cases is only presumptive, for the diagnosis could not be confirmed for the reasons already given.

The yearly incidence of surgical cases was as follows :—

1st year	16 cases
2nd year	31 cases
3rd year	35 cases
4th year	21 cases
5th year	40 cases
6th year	48 cases

Blair Aitken's figures for earlier years are : 1931, 7 cases ; 1932, 28 cases. In 1934, whilst I was in Lagos for a few months, a similar incidence of cases was observed. But at this time all cases of suspected peptic ulcer were operated upon and the abdomen explored. In the six years under review, a large but indeterminate number of cases have been treated in the medical wards. Even without allowing for these, the surgical figures show a steady increase in the numbers coming to hospital.

Out of 191 cases confirmed at operation in the six years, I have operated on 124 of them. The remaining 67 have been treated by surgeons working with me or who relieved me during periods of leave. It is on these 124 personal cases that the remainder of this paper is based.

Age Incidence.—The majority of these cases were between 30 and 40 years. Some were older and some were younger. As many of the older people were indefinite about their age, further inquiry into age groups was of doubtful value. But two cases were remarkable : these were two boys, aged 11 and 13 respectively, who presented themselves with well-established pyloric stenosis.

Sex Incidence.—Out of the 124 cases, 112 were males and 12 were females. (In Aitken's 48 cases, 44 were males and 4 were females.)

MORBID ANATOMY

The most striking feature of this series of cases was the distribution of the ulcers. 123 were duodenal ulcers and only 1 was a gastric ulcer, found in a male patient. In the 20–30 cases of peptic ulcer I saw in Nigeria previous to this series, there was only 1 case of gastric ulcer, also in a male. Both these gastric ulcers were situated on the lesser curvature of the stomach : one had penetrated through to the left lobe of the liver and the other had penetrated through to the pancreas.

Of the 123 cases of duodenal ulcer, 3 presented themselves as acute perforations—a fairly low rate of incidence.

The remaining 120 chronic duodenal ulcers showed a most striking pathological picture. The majority of these cases presented a greater or lesser degree of ' pyloric stenosis '. Hurst and Stewart (1929) have pointed out that the cause of pyloric stenosis as seen in Britain may be either a duodenal ulcer, a pyloric ulcer, or a pre-pyloric ulcer, and that it is sometimes difficult to say when the ulcer was originally in relation to the pylorus. In these cases there could be little doubt. The ulcer was situated on the anterior wall of the duodenum, often at an appreciable distance from the pylorus. On invaginating the stomach, the ring of the pyloric muscle could be made out and then a narrowing of the lumen of the duodenum beyond could be felt, distal and distinct from the pylorus. In the marked cases of obstruction, it was difficult to get the tip of the finger into the narrow lumen. But many cases of less degree of obstruction were apparent by the narrowing of the lumen. In the lesser degrees, it was difficult from this method of examination to say sometimes whether narrowing of the lumen was present or not. It is for that reason I have not given any definite figures for the actual number of cases of ' pyloric stenosis '. But in the lesser cases of obstruction, external examination of the duodenum showed scarring and puckering, which presupposed some narrowing of the lumen. In the cases of duodenal ulcer I have seen since my return to England, which cases have been of some standing, I have not observed an amount of scarring and fibrosis similar to that which these cases exhibited. In the advanced cases of obstruction the scarring and sclerosis were almost cartilaginous in consistency.

Thus the common feature of these duodenal ulcers was the relatively excessive formation of scar tissue, leading often to some degree of duodenal stenosis, loosely called pyloric stenosis. This form of pyloric stenosis is not sufficiently common in Britain for one person to see many cases, and I do not think this stenosis of the duodenum markedly distal to the pylorus is sufficiently appreciated.

The presence of this obstruction has a definite bearing on the clinical picture, which will be discussed later. In many of the cases, however, it is probable that there is spasm of the pylorus superadded to the organic obstruction, which helps to produce the symptoms of ' pyloric stenosis '. In many cases this spasm of the pylorus can be felt by the invaginating finger, the spasm relaxing gradually during the examination.

CLINICAL PICTURE

An analysis of the clinical picture as revealed by the questionnaire will now be given. This questionnaire was completed in the last 80 cases and the figures, therefore, relate only to the various symptoms and signs in these 80 cases.

Length of History.—This varied within wide limits.

11 cases gave histories of under 2 years.
50 cases had histories between 2 and 5 years.
11 cases had histories between 5 and 10 years.
8 cases had histories over 10 years, the longest being 20 years.

It will be noted that 61 out of 80 cases had histories of 5 years or under, a not unduly long history for duodenal ulcer in this country.

Presence of Intermissions.—Sixty-two cases gave a definite history of intermissions, of which 49 cases were free from symptoms between attacks and 13 had discomfort between attacks. Eighteen cases had continuous symptoms ; the majority of these were the patients with shorter histories. But the rather amazing finding was that several patients who showed marked visible peristalsis, and who, at operation,

were found to have marked duodenal stenosis with dilated, hypertrophied stomachs, also volunteered that they had periods of complete freedom from symptoms.

Many cases stated that recent attacks were longer and more severe. As this was elicited by rather a leading question, the value of this is doubtful.

Pain.—The ten headings under which pain is studied are taken from Ryle (1936), who has pointed out that these ten headings will elicit all the relevant facts about any pain.

1. *Character.*—Allowing for interpretations of different native languages, 52 described their pain as griping; others described it as pinching or burning.

2. *Severity.*—This is always difficult to judge. Forty-three cases said this pain was so severe as to stop work for the time being, when the pain was at its worst; 30 others said the pain was very severe. One interesting case said that he did not complain much of pain, but that his chief troubles were hiccupping and belching, with swelling of the abdomen. This man had a grossly dilated stomach, with waves of visible peristalsis like tropical ocean rollers.

3. *Site.*—The majority had the pain sited in the epigastrium, but 8 cases complained of the pain being chiefly in the right iliac fossa. One case complained of pain being in the left iliac fossa, and was misleading until examination eventually revealed visible gastric peristalsis, which made the diagnosis clear.

4, 5. *Paths of Radiation and Reference.*—It was difficult for the patients to differentiate between these, but 35 cases stated that their pain was not radiated or referred. The others stated that the pain was radiated to the back or iliac fossæ.

6. *Duration.*—The usual time of duration of the pain was 2–3 hours. Others said the pain lasted only half-an-hour, and a few said the pain lasted all day. The answers to this point did not seem very reliable.

7. *Frequency.*—The frequency of the pain varied from once to four times daily. This variation was interesting; on further questioning, a possible explanation was discovered in the different 'meal habits' of the various patients. Some had only one big meal a day. The most common habit was two big meals a day, with one or two snacks of 'agidi pap' (a thin starchy gruel, made from yams). The pain was usually found to be related to the big meals of the day, but in some patients was related to the taking of any food.

8. *Special Times of Occurrence.*—About half the cases complained of the pain waking them at night. As pointed out above, practically all cases related their pain with the taking of food, but the pain came at intervals varying from 10 minutes to 3 hours after food.

9. *Aggravating Factors.*—Sixty cases stated that their pain was aggravated by food.

10. *Relieving Factors.*—Only 4 cases stated that their pain was relieved by food; 63 stated that the pain was relieved by vomiting; 40 said the pain was relieved by resting; 32 said the pain was relieved by medicines, 'native' or otherwise, which medicines were presumed to be alkalis.

This analysis of the observations on pain calls for comment and will be discussed in the light of other findings later.

Vomiting.—Sixty-eight cases complained of vomiting, of whom 63 said the pain was relieved by vomiting; 22 cases induced vomiting to relieve the pain; 47 cases stated that the vomitus was large and foul-smelling. The remainder said the vomitus was small and usually clear.

In only 3 cases was blood observed in the vomitus.

But in some cases in which a very enlarged stomach was found, the patients did not volunteer any information about vomiting. Only on repeated questioning would they admit that they did vomit very occasionally, but obviously attached no importance to it.

Other Symptoms.—Belching was a common symptom. Appetite was usually reduced, particularly in the cases with visible peristalsis: some stated that they were frightened to eat because of the pain. The differentiation between loss of appetite and the fear of eating because of pain was very difficult.

One of the principal complaints was of the loss of weight.

General Appearances.—Many cases obviously confirmed the story of loss of weight. They were wasted, with sunken cheeks and a dry skin and in an extreme degree of under-nutrition.

Dental sepsis was not common.

Abdomen.—Fifty cases showed visible peristalsis. In several cases this had to be looked for on repeated occasions before it was discovered, but in many it was immediately obvious. Seventeen cases had such grossly enlarged stomachs that the peristalsis stood out like ocean waves. In 7 of these cases the enlarged stomach stretched down to the brim of the pelvis, filling the abdomen like a rugby football; one of these was the boy aged 13.

Succussion splash could be elicited in many cases that did not at first exhibit visible peristalsis.

Fractional Test-meal Findings.—This test was done on every case and the findings are interesting. Almost universally, there was a large volume of fasting juice, sometimes as high as 1000 c.c. There was a general increase in the amount of mucus present. Starch was always present and bile was never present up to the end of the 2½ hour test.

In the cases that showed visible peristalsis, the level of free HCl was low, with a relatively high level of total acidity.

In other cases, the level of free HCl was variable, but usually within normal limits. In three cases it was very high and one of these cases will be described in detail later.

General Discussion.—This analysis of symptoms and signs of a series of 80 consecutive cases of chronic duodenal ulcer gives a picture of the general way in which these patients present themselves to hospital for treatment. It will be observed that the classical picture of duodenal ulcer is very rare; 4 cases only complained of hunger pain. But as pointed out in the description of the morbid anatomy, most of the cases are complicated by some degree of obstruction. A consideration of individual clinical pictures, together with the morbid anatomical finding in each, reveals a very interesting picture of

a graded series of symptoms to correspond to the pathological state.

Four type cases illustrating landmarks in the changing picture can be chosen :—

1. Duodenal ulcer without obstruction.
2. Duodenal ulcer with beginning obstruction.
3. Well-established 'pyloric' obstruction with dilated, hypertrophied stomach.
4. Long-continued obstruction, in which a 'live and let live' compromise appears to have been reached.

It is the fourth type of case which is most interesting and which I have not seen well described before. It is exemplified by the man already referred to, who complained of no pain or vomiting, but whose chief troubles were hiccupping and belching and a dilated stomach. I have mentioned other cases with very enlarged stomachs where vomiting was not at all a prominent feature.

The stomach in these cases appeared to have given up the unequal struggle to force food through into the duodenum in the normal way. It was now merely a reservoir of ingested food, which digested and fermented, and it so tolerated this fermenting mass that it did not even bother to eject it. These cases are a stage beyond the normal state of affairs in pyloric stenosis and show the adaptability of the stomach to abnormal conditions.

The first and third types are the usual extremes as seen in Britain of duodenal ulcer with its complications.

The main findings in type 1 are :—

a. Pain a long time after food and relieved by food ; *b*. Remissions ; *c*. Pain waking the patient at night ; *d*. No vomiting or the vomiting of small quantities of clear fluid ; *e*. Relatively small quantity of fasting juice in the test-meal ; *f*. No visible peristalsis ; *g*. No loss of weight and a fair appetite.

The main findings of type 3 are :—

a. Pain a short time after food and aggravated by food ; *b*. No remissions ; *c*. Pain not waking the patient at night ; *d*. Vomiting of large quantities of foul fluid ; *e*. Large quantity of fasting juice in the test-meal ; *f*. Visible peristalsis ; *g*. Loss of weight and a poor appetite.

In type 2, between these extremes, there is to be seen a gradual change of symptoms to correspond with an increasing degree of stenosis. It is among these patients chiefly that 22 cases who induced vomiting to relieve the pain are to be found.

The connexion between observed visible peristalsis and the degree of stenosis found at operation was somewhat variable. In two cases when scarring and the amount of stenosis appreciated by the invaginating finger was similar, one had shown peristalsis and one had shown no peristalsis. In the discussion of the morbid anatomy, I have already referred to the spasm of the pylorus which may be present, in addition to the organic stenosis, and it is probably this which is the real cause of calling forth violent peristalsis, which becomes visible. Normal gastric peristalsis is sufficient to overcome a slight degree of organic stenosis, but some dietary indiscretion produces spasm and violent peristalsis is required to overcome it. Visible peristalsis may, therefore, be an inconstant feature, and it may occur in cases even though repeated observation in hospital fails to discover it. The following case illustrates this :—

A man, aged 35, was admitted to the medical wards complaining of dyspepsia. He was treated by diet for three weeks and at the end of that time was discharged free from symptoms. Doubtless he went out and had a large spiced meal as a change from his monotonous therapeutic diet. But at the end of a week he returned to surgical out-patients showing most marked visible peristalsis. At operation an appreciable amount of scarring with narrowing of the lumen of the duodenum was found.

TREATMENT

Operation Results.—All the 120 chronic cases were treated by gastro-enterostomy. All of them, except for the two young boys, aged 11 and 13, were operated upon under spinal anæsthetic (light percaine). There was only 1 death in the series. This was the boy aged 13, who was found to have the ulcer perforated some time just prior to coming to the operating theatre for a gastro-enterostomy. The perforation was sutured and the gastro-enterostomy was done. He did very well for ten days after the operation and then died suddenly. Unfortunately the parents refused permission for a post-mortem and the cause of death was not definitely ascertained.

Post-operative History.—

Immediate.—The post-operation régime of these cases is given in the appendix. The diet described was devised in an endeavour as far as possible to use local foodstuffs to which the patients were accustomed. It has proved very successful in practice. The patients stayed in bed for fourteen days after operation and were discharged to out-patients a few days later. Except for the one death, the patients' convalescence gave rise to no anxiety and the trivial constitutional disturbance produced by the operation was very remarkable.

Later History.—The patients were weighed on discharge from hospital. Their progress the first few weeks after discharge was most astonishing. The weight increased rapidly, many cases putting on a stone in two weeks. The sunken cheeks filled out and the patient grew sleek and in some cases became almost fat. Of course, those who showed the most change were those who had the most stenosis.

Post-operative Complications.—A follow-up clinic was instituted and most cases attended hospital to report for at least six months after operation and many cases have been followed for much longer. One interesting sequel to operation was the swelling and œdema of the feet and ankles that often developed shortly after patients began walking about again. This œdema was never present before operation. The few blood examinations that were done before and after operation gave no clue to the cause. These cases were treated purely empirically with a simple iron mixture and the œdema cleared up in three or four weeks.

Apart from the above, and one particularly interesting case about to be described in detail, late complications were conspicuous by their absence. The patients continued to be free from pain and vomiting. They were able to return to work and resume their normal diet.

The one case in which an anastomotic ulcer did develop was as follows :—

The patient, a man aged about 30 years, was operated upon in September, 1940. He had a history of two years' dyspepsia, very occasional vomiting, but not appreciable loss of weight. A test-meal showed a high level of free HCl. At operation, a duodenal ulcer without stenosis was found and a gastro-enterostomy was performed. The usual uneventful post-operative course followed and after a few routine visits to the follow-up clinic, the patient disappeared. In June, 1943, the patient returned complaining of similar symptoms, which he said had been present for six months. Vomiting was now more frequent and he had lost a little weight. The test-meal showed a very much lowered free HCl and no bile was found in the gastric juice. A diagnosis of stomal ulcer was made and he was put on the peptic ulcer diet in use in the medical wards. After a few days on the diet, it was suddenly discovered that visible peristalsis was present; this may have been missed before, because we did not look carefully for it. The diet was continued for some time, but the visible peristalsis persisted. The abdomen was, therefore, opened again. A large stomal ulcer was found; the stoma admitted two fingers. In addition a marked degree of pyloric stenosis was present due to almost cartilaginous scarring of the original ulcer. A second, anterior gastro-enterostomy was performed. The patient had a normal convalescence and was discharged from hospital on the sixteenth day. The fifth day after discharge, the patient returned with all the signs of a perforated ulcer. On opening the abdomen, a perforation of the jejunum ¼ in. distal to the recent anastomosis was found; the perforation was quite distinct from the anastomosis. The ulcer was sutured and the patient again had an uneventful convalescence and since discharge has been free from symptoms.

This history presents several interesting features :—

1. The presence of chronic obstruction in the stomach as revealed by visible peristalsis, in spite of a second escape route, two fingers wide. It would appear that the stomal ulcer caused enough spasm to prevent the stoma from working. The absence of bile in the gastric juice showed that regurgitation into the stomach was also prevented. The marked diminution in gastric acidity must have been due to some other cause than the regurgitation of alkaline jejunal contents.

2. The continuation of the pathological process in the duodenum with the production of stenosis. Hurst and Stewart suggest the treatment of stomal ulcer is to unpick the anastomosis and excise the ulcer because the original ulcer is usually healed.

How long is required for stenosis to be produced in a duodenal ulcer is a matter for speculation, but in this man it occurred in two years.

3. The rapid onset of the third, the perforated ulcer, which required 21 days from start to perforation, is of great interest. Hurst and Stewart record a similar case of multiple ulcers in a case where two anastomoses had been performed.

This one case of ill-result of treatment, together with only 1 death in 120 cases, is most gratifying. One possible cause of the good results of operation in this series may be found in the results of the test-meals. As already pointed out, the level of free HCl in most cases was low. In one of the few cases where the level was high, a stomal ulcer developed, as described above. The highly acid gastric juice is regarded as one of the potent factors in the production of stomal ulcers and in the majority of the cases in the series the jejunum did not have to contend with this. This series of cases also serves to emphasize what a useful operation gastro-enterostomy is in the proper case.

AETIOLOGY AND PATHOGENESIS

The discussion of this has been left till the last for there are many interesting facts for study and speculation.

The proportion of 123 duodenal ulcers to one gastric ulcer in this series suggests a different exciting cause for the two types of ulcer.

We had one example of familial incidence. One man who had been operated upon two years before, walked in fat and well about a year ago, bringing his son aged 11 for operation.

Now Maingot suggests that the incidence of duodenal ulcer is directly related to the exigencies of modern civilization. Boland (1942), writing about a hospital in the Southern States of the U.S.A., points out that the incidence of duodenal ulcer is much larger in the white population than in the black. He further goes on to state that the incidence in the black race is slowly increasing. He suggests that this is due to the fact that the black race is abandoning its own native diet and is now consuming the pernicious diet of civilization. But my cases almost without exception are people living remote from the hurly-burly of civilization and consuming their native diet. Somervell and Orr (1936) produced some very interesting facts about the incidence of duodenal ulcer in India, which facts, in my opinion, have not been sufficiently appreciated. They pointed out that in some areas ulcer was very common, that in others, geographically near, ulcer was very rare. The main difference appeared to be that in the non-ulcer area rice was a main part of the diet and in the ulcer area it was not. The diet, therefore, not necessarily of civilization, has an important bearing on the incidence of ulcer. In my cases rice was not a staple of diet, but usually formed some part of it. The cases came from areas up to 100 miles away from Lagos and as far as I was able to discover, there were no radical differences in diet in any of these areas.

A fair case can be put to make a dietetic factor, possibly a vitamin, the villain of the piece in the causation. In the native races, there is an absolute lack of vitamin. In the white races, the methods of preparing or preserving food may destroy the factor, or else the hectic life, with its hurried, irregular meals, may prevent its proper absorption.

To speculate on the nature of this factor is invidious. But one negative contribution may be given. It is popularly supposed in Nigeria that peppers may be an exciting cause. In fact red and green peppers are ingested in very large amounts in the native diet. But if peppers were an exciting cause, peptic ulcer would be a very much commoner disease than I have shown it to be. In my opinion, therefore, they are not important as an exciting cause. I think, however, that they may be a factor in the pathological picture. Once an ulcer is present, the gross irritation of these fiery peppers may cause the excessive formation of fibrous tissue and the scarring which is present in the large majority of cases.

SUMMARY

1. The incidence of peptic ulcer in a six-year period at the African Hospital, Lagos, Nigeria, is described and is shown to be increasing. It is probable that this is due to an increase of cases diagnosed coming to the hospital, rather than to increase in the rate of occurrence.

2. 124 personal cases of ulcer are described and the age, sex, and anatomical distribution is discussed.

3. The clinical picture of the last 80 of these chronic cases is analysed. The almost universal presence of some degree of duodenal obstruction, due to excessive fibrosis, is recognized. The modification of the clinical picture to correspond with the pathological change is discussed.

4. The treatment of these cases is described. The satisfactory results of treatment are given and the reason for them are discussed.

5. A brief discussion of the pathogenesis of peptic ulcer is given, and reference made to factors familiar to the cases described.

I am indebted to the Honourable The Director of Medical Services, Nigeria, for permission to publish this paper.

APPENDIX I

POST-OPERATION DIET OF GASTRO-ENTEROSTOMY CASES

Rectal saline 1 pint immediately after return from operation theatre: thereafter ½ pint 4-hourly.

1st day after operation: Rectal saline ½ pint 4-hourly. 1 oz. sterile water by mouth every 2 hours.

2nd day: 2 oz. sterile water by mouth every 2 hours. Every 4 hours 1 oz. of milk to be added to the water.

3rd day: 3 oz. of sterile water every 2 hours. Every 4 hours 2 oz. of milk to be added to the water.

4th day: Sips of water in moderation. 2 oz. of milk every 2 hours, except at midday when 4 oz. of egg-custard to be given.

5th day: Water in moderation. Agidi pap 2 oz. at 8 a.m. Custard at midday.

6th *day:* Agidi pap 3 oz. at 8 a.m. Custard 4 oz. at midday. Cornflour 3 oz. at 4 p.m. Every other 2 hours, milk 4 oz.

7th day: Agidi pap 3 oz. at 8 a.m. Rice 2 oz. at noon. Cornflour 3 oz. at 4 p.m. Every other 2 hours, milk 6 oz.

8th day: Agidi pap 3 oz. at 8 a.m. Rice with 4 oz. plain soup, at midday. Cornflour 3 oz., 4 p.m. Every other 2 hours, milk 6 oz.

9th day: As for 8th day.

10th day: As for 8th day.

11th day: Agidi pap 3 oz. at 8 a.m. Rice 6 oz. with plain soup at midday. Rice 6 oz. with plain soup at 4 p.m. Every other 2 hours, milk 6 oz. And so on until discharge.

NOTES:—

1. Agidi pap is a kind of gruel made from yams, with a little sugar added. It is a staple form of light meal in Southern Nigeria.
2. Cornflour is locally produced from ground maize and cooked into a pudding with milk.
3. Plain soup is made from meat or fish and green vegetables, the latter consisting of local herbs. It is more like a thin stew. It is called 'plain' because peppers, red or green, are not added. Most native foods are highly flavoured with these peppers and the ulcer cases are deprived of these strong irritants.

APPENDIX II

QUESTIONNAIRE

Name Hospital No. Date
Age
Complaint
Previous History
Duration and progress of Symptoms:
1. Short History.
2. Long History:—
   a. With continuous symptoms.
   b. With intermissions:—
      1. Free from symptoms.
      2. Discomfort between attacks.
      3. Almost continual mild symptoms with periodic exacerbations.

Description of attacks and intervals.
a. Short / Long attacks with short / long intervals
b. Recent attacks becoming longer and more severe.

Pain:—
- Character
- Severity
- Situation
- Radiation
- Paths of reference
- Duration
- Frequency
- Special times of occurrence
- Aggravating factors
- Relieving factors

Vomiting:—
- Absent
- Frequent
- Continuous
- Colour
- Odour
- Fæculent
- Coffee grounds
- Blood
- Other factors
- Large
- Small
- Relation to food
- Relation to onset of pain
- Self-induced
- Relief of pain

Nausea; regurgitation; flatulence; belching; sense of fullness; heartburn.
Weight: rapid loss; gradual loss; stationary; gaining.
Appetite: normal; anorexia; diminished for fear of pain; increased.
Hæmatemesis and melæna:—
- Frequency
- Quantity

With or without collapse

Jaundice:—
- Number of attacks
- Preceded by pain or colic with/without pyrexia
- Depth
- Dark urine or light stool noticed

Condition of bowels:—
- Normal
- Constipated
- Diarrhœa
- Other features.

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ATYPICAL CROHN'S DISEASE

A REPORT OF THREE CASES

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SINCE Crohn, Ginsberg, and Oppenheimer published a paper based on 14 cases of regional ileitis in 1932, much has been written about these cases, especially in America, where by 1937 over 200 cases had been recorded. Crohn's original description of the disease has been found to apply to the majority of cases. Usually the disease is maximal at the termination of the ileum, gradually diminishing in a proximal direction so that the wall of the ileum becomes gradually normal at about 10 to 14 in. from the ileocæcal junction. The main modification of the original description was that the cæcum and the ascending colon may also be affected. Most surgeons who deal with abdominal emergencies have encountered the inflammatory phase which causes the symptoms and signs suggesting appendicitis: it is thought that resolution can occur at this stage because many cases, diagnosed by laparotomy only, have remained symptom-free for a year. The obstructive phase of the disease is less common and it may be associated with internal or external fistulæ; a gross deterioration in the general condition is the rule in these cases. We have not encountered or recognized the type with diarrhœa, suggesting enteritis, apart from the formation of an internal fistula and the alternate diarrhœa and constipation associated with the intestinal obstruction.

A. W. Taylor (1945), in contradistinction to Crohn and Hadfield, thought that this condition has had a far too enthusiastic reception as a new disease; he considered that it is impossible to distinguish Crohn's disease from hypertrophic ileocæcal tuberculosis by means of clinical features or morbid anatomy. In his three cases, two of which died in a few months from miliary tuberculosis, tubercle bacilli were found in the resected specimens which simulated Crohn's disease. In the three cases about to be presented tuberculosis was excluded as far as possible by Ziehl Neelson stained sections and culture of histological tissue. No evidence of any acid-fast bacilli was found. In the first two cases the typical changes associated with this disease were found in the terminal ileum and in the third case identical changes were found in the cæcal wall.

From the pathological point of view the disease is not usually difficult to diagnose: generally it conforms to the classical description of the disease by Hadfield (1939) and the diagnosis is made mainly on the macroscopical distribution and the type of lesion, together with a histological picture of chronic non-specific inflammation with the exclusion of tuberculosis as far as possible. Obviously if atypical lesions are to be found in this disease, then difficulties of diagnosis will arise, but it will be seen from these three cases that although two of them were unusual from a pathological point of view macroscopically, microscopically they conform to type.

CASE REPORTS

Case 1.—Private T., aged 18, was admitted on Feb. 3, 1947, with a three months' history of slight intermittent abdominal pain. This pain was central, diffuse, and colicky, while in the last two weeks he had a more constant pain in the right side of his abdomen. He had a tendency towards constipation, but there was no nausea and no vomiting had occurred. There were no urinary symptoms.

ON ADMISSION.—His temperature was 99·2° F., pulse-rate 90 per minute, and his tongue was furred. A large mass was palpable to the right of the umbilicus; there was little tenderness, no rigidity, and the mass could be pushed forwards from the right loin.

It was considered that he probably had an appendix abscess although the mass was much higher and more central than the usual appendix mass. His urine and an intravenous pyelogram were normal, but he had 24,000 white cells per c.mm. of blood, with 78 per cent of polymorphonuclear cells. He was about to have a barium meal when on Feb. 9, 1947, he developed severe pain in the right hypochondrium associated with much more tenderness and rigidity.

AT OPERATION.—The omentum, covered with fibrin, was found to be adherent to a large mass of mesenteric glands in the ileocæcal angle. A probe could enter the mass at one area fairly close to the ascending colon and a small amount of odourless pus was exuding from the mass. There was no direct communication with the lumen of the bowel and later the pus was found to be sterile. The terminal ileum showed the typical macroscopical changes of Crohn's disease and the disease had apparently spread to the cæcum, but the appendix was scarcely affected. The middle of the transverse colon was also attached to the mesenteric mass, so that the resection included about 10 in. of affected ileum with the right half of the colon. The main difficulty at the operation was the removal of the mesenteric mass without injuring the adherent ureter: a few of the smaller glands were left behind.

PROGRESS.—His immediate recovery was excellent, but on Feb. 24, pyrexia and abdominal pain recurred: two days later temporary relief followed a purulent discharge from the old drainage stab wound in the right loin; this pus was found to contain staphylococci and *B. coli*, but no evidence of tuberculosis was found by film or culture on several occasions. He was still at this time suffering from anorexia and loss of weight. A few of the remaining mesenteric glands were palpable per rectum high up on the right side.

By March 19 there was marked clinical improvement, he was eating well, gaining weight, and there was little discharge from the loin; the mesenteric glands palpable per rectum were much smaller and on discharge from hospital in April the glands had disappeared completely. Since discharge from hospital he has carried out his work as a male nurse without any further symptoms or trouble.

PATHOLOGICAL REPORT (*Fig.* 96).—Macroscopically the specimen consists of the terminal 10 in. of the ileum, the cæcum, the ascending and transverse colon surrounding a hard mass enclosed in the ileocæcal angle and adherent to the transverse colon. Anteriorly the omentum, adherent to the mass over an area about 4 in. square, can be readily separated and pus can be seen exuding from a small hole on the anterior surface of this mass. There are numerous small discrete lymphatic glands to be seen in the mesentery of the ileum, medial to this mass. The appendix, in the retrocæcal position, appears normal.

On hemisection of the specimen it is observed that the walls of the ileum and the lower part of the cæcum are grossly thickened, being mainly $\frac{1}{2}$–$\frac{3}{4}$ in. in thickness. The thickened walls have a whitish, fibrous, homogeneous appearance: the mucous membrane of the ileum is hæmorrhagic, though not ulcerated macroscopically, and has rather a rough appearance. The thickening in the wall of the cæcum is entirely in the portion below the entrance of the ileum and is seen to involve the ileocæcal valve.

The mass in the ileocæcal angle is about $3 \times 2\frac{1}{2} \times 2$ in., firm and rather rubbery in consistency. It has a whitish, mottled appearance, with two areas of necrosis in the centre, one of which communicates with a hole in its anterior surface. This mass, presumably of lymphatic glands, is intimately adherent to the terminal 2 in. of the ileum and to about 1 in. of the ascending colon around the ileocæcal angle; it is also adherent to the right half of the transverse colon. The mucous membrane of the upper part of the cæcum and the colon appears normal.

Histology.—

1. *Ileum*: The main changes of lymphœdema and round-cell infiltration occur in the submucosal layer, although they are present to a lesser extent in the other layers of the gut. Fibrosis is marked and long-standing.

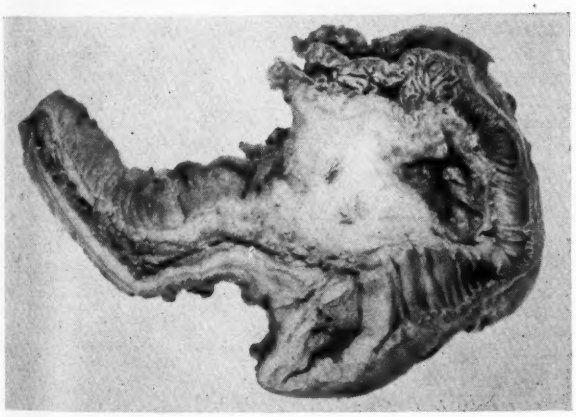


FIG. 96.—*Case* 1. Bisected specimen showing typical regional ileitis with a large mass of necrotic but non-caseating mesenteric glands. (Specimen now in the Millbank Museum.)

The mucous membrane is acutely ulcerated microscopically in some areas, there being small localized areas of inflammation extending into the inner half of the thickened submucosal layer.

2. *Glands*: The section consists of a small localized area of inflammatory cells surrounded by areas of fibrosis. The inflammatory reaction consists mainly of plasma cells and fewer lymphocytes with occasional polymorphonuclears. Irregular giant cells are seen in some areas with peripheral nuclei. Fibrous tissue, both young and old in type, is present in large amounts and no lymphatic gland structure is left. In the centre of the cellular areas polymorphonuclear cells predominate. One small area of necrosis can be seen containing many extravasated red blood-corpuscles. No caseation was seen.

3. *Bacteriological examinations*: The 'pus' and tissue were cultured and smears were examined for tuberculosis. In addition numerous stained sections were examined for acid-fast bacilli, but none were found.

Case 2.—Private W., aged 22, first suffered from attacks of anorexia and vomiting in April, 1944. In September, 1944, attacks of abdominal colic associated with vomiting occurred, but he did not seek medical advice until December, 1944, when he was off work with a diagnosis of debility and gastritis for four months. In April, 1945, he resumed work, but he often had days off with his previous symptoms. His abdominal pain was relieved by the onset of diarrhœa half an hour after meals. In November, 1945, he started to have pain behind the right knee, spreading up to the right buttock. At this time his abdominal symptoms were less marked apart from persistent diarrhœa. His doctor sent him to a Tuberculosis Officer because he found a swelling in the right buttock, but the patient returned with a 'negative report'. He was therefore sent to a provincial teaching hospital where medical treatment for his sciatica was advised. In February, 1946, after a negative X-ray examination of his hip, he was ordered massage and ultra-violet light at another hospital. He was called to a Medical Board examination in April, 1946, and a month later was placed in Category B.1. He was called up for military service in August, 1946, but he reported sick with sciatica after one week: a month later his sciatica was worse and associated with abdominal pain, so he was sent to hospital.

ON EXAMINATION.—He was first regarded as a case of peri-arthritis of the right hip-joint as the X-ray examination of this joint was still negative, but on re-examination of the films gas could be seen in the right buttock. His general condition was obviously deteriorating, with intermittent pyrexia, and the elusive swelling of the right buttock appeared for a short time in October, 1946. The swelling reappeared in December, 1946, when 30 c.c. of foul pus were aspirated, which grew *B. coli*. A rectal examination revealed a pelvic abscess, which burst on the removal of the finger. His general condition, i.e., pyrexia, emaciation, and anorexia, failed to improve much even with the aid of blood transfusions and vitamins, also the swelling of the buttock reappeared. The result of a barium meal examination was disappointing; it merely showed a doubtful filling defect in the ileocæcal region, but no definite 'string sign': no barium entered the buttock. A barium enema showed a possible 'string sign', but the appendix was not visible.

AT OPERATION.—On Feb. 3, 1947, laparotomy showed the presence of an inflammatory mass in the pelvis; the appendix tip was buried, but the proximal part was dilated and thickened. A loop of the terminal ileum was entering and leaving the pelvic mass and the proximal loop was dilated with normal thin walls: the efferent loop from the mass to the ileocæcal junction was thickened, contracted, and œdematous, with little lumen. Owing to the poor condition of the patient a lateral anastomosis between the afferent and the efferent loops was performed after removing the base of the appendix.

PROGRESS.—He had a stormy recovery and a fæcal discharge from the stab drainage in the right iliac fossa developed two days after the operation. On Feb. 11 the intermittent buttock swelling was drained and the communication with the pelvis through the great sacro-sciatic foramen was confirmed on March 21. His condition then improved and he gained some weight; he could, however, eat only two meals a day, otherwise he developed abdominal pain. A barium meal at this stage showed no obvious intestinal obstruction but the colon loaded with fæces appeared to be by-passed: again no barium entered the buttock.

SECOND OPERATION (May 5).—The previous anastomosis appeared to be working because the ileum proximal to it was not dilated but the portion between the anastomosis and the pelvic mass was dilated. The efferent

terminal ileum presented as before the typical appearance of Crohn's disease. The indurated abscess cavity mainly on the right side of the pelvis had no communication with the appendix stump because this was later proved to

FIG. 97.—*Case* 2. Photograph showing condition just before the second operation. His emaciation has improved and he has two discharging sinuses in the buttock.

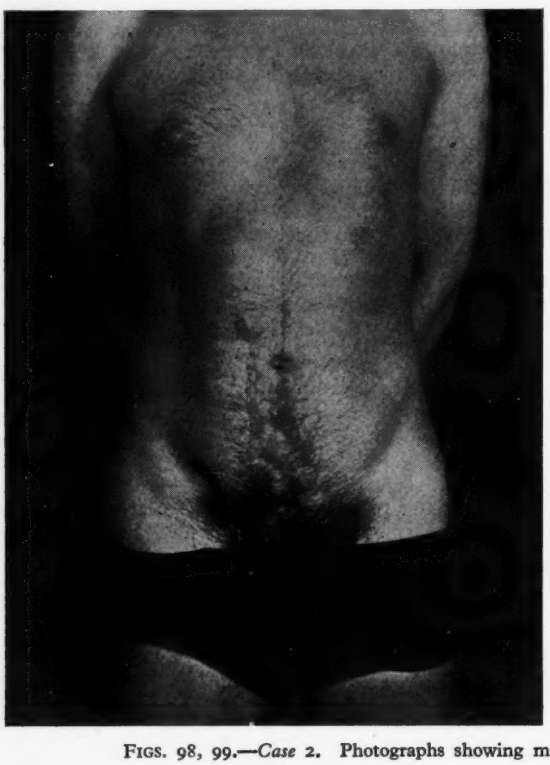
be closed by examination of the specimen. The whole of the affected 2 ft. of terminal ileum and the right half of the colon were resected with end-to-end anastomosis: a fistula into the rectum was also closed. It was noted at this operation that the colon was loaded, in spite of an empty rectum with a previous liability to diarrhœa. Pre-operative enemata had failed to remove these scybala, which may be explained by the fact that on one occasion part of the enema was expelled through the buttock!

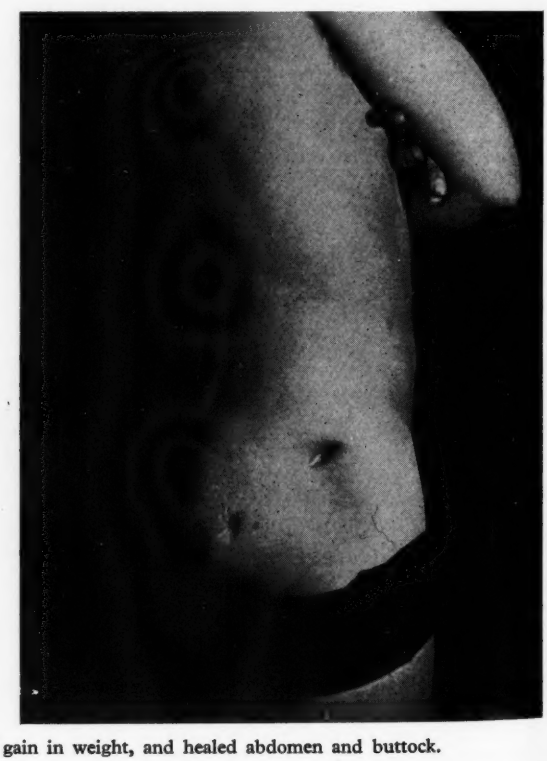
PROGRESS.—His rapid recovery from this operation was amazing after the initial two days: the buttock sinuses closed and abdominal symptoms ceased. On May 19 he had fæcal impaction in the rectum requiring manual removal. By June 7 he was free of sciatica and had no discharge from his abdominal wounds or buttock; he was gaining weight at the rate of 1½ lb. per week and enjoying his food without abdominal pain for the first time for three years.

Progress after discharge from the army has been maintained apart from a temporary attack of hepatitis with jaundice in the latter part of July: this was almost certainly due to a plasma transfusion. He has resumed his work as an electrician. In December he wrote to say that he was extremely well and 12 st. 4 lb. in weight. (*Figs.* 98, 99.)

PATHOLOGICAL REPORT (*Fig.* 100).—Macroscopically the colon and the majority of the cæcum appear normal. Externally the inferior aspect of the cæcum appears thickened and there is an appendix stump rough on the outside. The terminal ileum is in the form of a loop, the two ends of which are joined about 2 in. from the ileocæcal junction by an ileo-ileostomy. There is a fæcal fistula present in the most dependent part of the loop about 6 in. from the ileocæcal junction.

On longitudinal section the colon is normal, containing a large, firm mass of scybalous fæces. The cæcum contains a large mass of hard fæces. It is quite normal apart from the lower portion, which shows thickening of the wall, particularly around the appendix stump, the wall of this area being ½–¾ in. thick, firm, and hæmorrhagic looking. No opening could be found between the cæcum and the appendix stump. About 14 in. of ileum are present in the form of a U-shaped loop. The ileo-ileostomy is patent and well healed.





FIGS. 98, 99.—*Case* 2. Photographs showing marked gain in weight, and healed abdomen and buttock.

No ulcers are seen in the bowel apart from the area leading to the fistula. The mucous membrane has, however, a rough mosaic type of appearance. For the first 6 in. the wall of the ileum is thickened and firm, being ¼–¾ in. in thickness. The lumen of the gut is difficult to find and is completely occluded. About 6½ in. from the ileocæcal valve there is a fistula, which communicates with the exterior of the bowel. The gut gradually returns to normal after this point, until normal gut is seen 10 in. from the ileocæcal valve.

Histology.—

1. *Cæcum and Appendix Stump :* The cæcal wall is apparently normal apart from a small amount of cellular infiltration and a fair amount of œdema particularly in the muscle and submucosal layers. The mucous membrane is normal. The area around the appendix stump consists of a subacute inflammatory reaction with numerous polymorphonuclears.

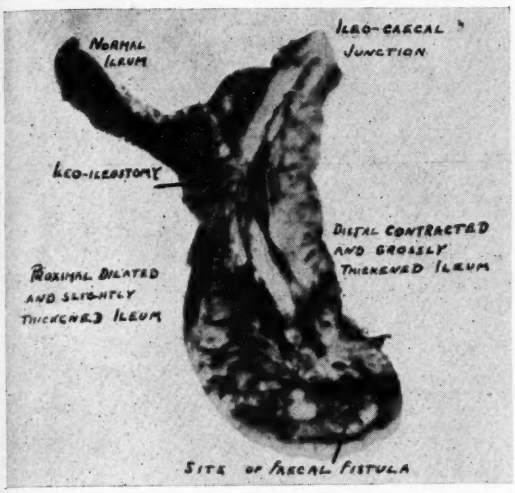


FIG. 100.—*Case* 2. Photograph showing the resected loop of ileum and the previously made ileo-ileostomy. The resected right half of the colon has been detached.

2. *Ileum* (2 in. from the ileocæcal valve) : This specimen shows lymphœdema, particularly of the submucosal layer, with considerable round-cell infiltration throughout. Normal structure of the part is maintained, but a fair fibrous tissue reaction is present.

3. *Ileum* (4 in. from the ileocæcal valve) : This specimen shows the same changes as above, together with a small pyæmic abscess in the wall in relation to an ulcerated area.

4. *Bacteriological Examinations :* Culture and smears of tissue were all negative for tuberculosis.

No giant cells and no caseation were seen throughout. The cellular reaction was predominately lymphocytic and plasma cell with fibrous tissue reaction fairly advanced, particularly in the submucosal layer. Lymphœdema was marked and ulceration minimal.

Case 3.—A woman aged 45 years, who was a theatre sister before she was married. A few months before admission she had an attack of abdominal pain and an X-ray examination at the Manchester Royal Infirmary showed that gall-stones were present. No surgical treatment was performed because she had only a short spell of mild symptoms. While on holiday she developed an attack of central abdominal pain, which settled in the right iliac fossa.

ON ADMISSION.—She was admitted on June 1, 1939, on the third day of the attack, with a temperature of 99·7° F. and a pulse-rate of 100 per minute. Her tongue was furred and there was tenderness with rigidity in the right iliac fossa.

AT OPERATION.—Under anæsthesia a mass in the right iliac fossa was palpable. A small quantity of odourless pus was present in the peritoneal cavity in the region of the mass, to which the omentum was adherent, present on the inner side of the ascending colon extending to the posterior abdominal wall and mesentery. The appendix and ileum were normal. The right half of the colon and about 5 in. of normal terminal ileum were resected with end-to-side anastomosis of the ileum into the transverse colon and closure of the distal end of the transverse colon. The gall-bladder was not thickened or inflamed but as it contained two gall-stones it was removed. It was also noticed that the uterus was enlarged, corresponding to a nine-weeks' pregnancy.

She made an uneventful recovery except that three days after operation she had two days of vaginal bleeding : pregnancy, however, proceeded, but she had to have a Cæsarean section at full term and now has a healthy daughter just eight years old. She was readmitted in June, 1944, with some right-sided abdominal pain and frequency which turned out to be a *B. coli* pyelitis. An intravenous pyelogram showed no abnormality. A barium meal and enema were also arranged and there was no sign of any recurrence of Crohn's disease.

PATHOLOGICAL REPORT (Professor T. B. Davie, June 10).—"The affected part of the cæcum shows intense mucosal and submucosal œdema together with some acute non-suppurative inflammation of the mucosa. The thickened pericæcal tissue shows masses of subserous fat infiltrating with the cells of subacute inflammation. In several places there are acute abscesses. There are no signs of tuberculosis, actinomycosis, or malignancy and the condition appears to be an acute phase of Crohn's disease mainly affecting the cæcum."

DISCUSSION

The first case is atypical owing to the size of the mesenteric glandular enlargement, but in this case the mass of glands was so large that the centre was necrotic and the bursting of the affected mass was the cause of the symptoms of peritonitis. The only case with 'enormous' glands that we have found was described by Jackson (1937).

The clinical history of the second case may be unique. One curious feature was the repeated evanescent swelling of the right buttock so that, with a history of sciatica, emaciation, pyrexia, and the presence of slight limitation of the hip movements, the diagnosis of a tuberculous hip was probable. The swelling subsided while preparations were being made to aspirate or drain it, so that this case had perplexed many competent physicians, orthopædic surgeons and general surgeons for a long time. Another difficulty in diagnosis was that the patient had minimized his abdominal symptoms when the sciatica appeared, so that attention was not focused on the alimentary tract until foul pus was aspirated. The diagnosis was not even obvious at the first operation because the tip of the appendix was involved in the inflammatory mass and the base of the appendix was affected. This case also shows that resection is preferable to short circuit with exclusion, but this patient would have probably succumbed to primary resection. It is possible that he might have improved more rapidly after the first operation if the dilated proximal ileum had been anastomosed to the transverse colon, but this would have left an even longer length of ileum between the anastomosis and the site of the intestinal obstruction and fistulæ. According to Dickson Wright an end-to-side anastomosis between the ileum and the transverse colon, with closure of the distal end of the ileum, is a very satisfactory procedure for Crohn's disease : he doubts whether resection, with its associated mortality, is necessary even when fistulæ are present. I did not consider this method at the time because it would have meant closing a dilated ileal stump invisibly obstructed in a pelvic mass. This anxiety could have been overcome by bringing the ileal stump to the surface of the abdominal wall as is often

performed during a terminal ileostomy for ulcerative colitis.

The most unusual feature of the third case is that the disease affected the cæcum without the terminal ileum. Only a small piece of terminal ileum was removed to enable anastomosis to be performed. This patient has lived for eight and a half years after the operation and she is in good health. She was seen three years ago with an attack of pyelitis and the opportunity of a barium meal examination showed no evidence of recurrence.

We are indebted to many colleagues for the successful outcome of these cases, especially to the anæsthetists Lt. Col. Scriven, M.B.E., Major Stevens, and Dr. Pugh Thomas. Lt. Col. Ettles gave invaluable help and advice. We also wish to thank Brigadier Fettes, O.B.E., Consultant Surgeon to the Army, for permission to publish the two military cases.

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CARCINOMA OF THE ŒSOPHAGUS: A CLINICO-PATHOLOGICAL STUDY

BY RONALD W. RAVEN, O.B.E., LONDON

THE development of the surgical treatment of carcinoma of the œsophagus constitutes one of the most important advances in surgery during the present decade, and illustrates the reward of tenacity of purpose in a difficult field. For many years interest in the subject was maintained by the publication, at long intervals, of an occasional case of successful surgical operation, and there were those who looked to the future with confidence and hope. During this preparatory period anatomical problems were dealt with and the various methods of surgical exposure of the œsophagus were established. Attention was also directed to the pathology of the disease, and it was necessary to learn more about the early symptomatology, so that the diagnosis could be established during the early phase of development.

This study was undertaken to elucidate certain problems in connexion with the pathology of the disease when patients reach hospital for treatment; to learn more of the early symptomatology; and to determine the duration of survival after all methods of treatment, excluding surgical excision. A knowledge of the pathological state of these patients will enable the operable cases to be differentiated from the inoperable, and is necessary if satisfactory results are to be achieved by radical operation. It is an unfortunate fact that so large a proportion of patients come to hospital when the disease is far advanced, and radical surgery is thus contra-indicated. A better understanding of the early symptomatology is required so that the diagnosis can be made during the initial phases of the disease and more patients will undergo radical surgical excision.

The basis of this paper is a series of 277 patients who were admitted to the Royal Cancer Hospital and have been studied from the clinico-pathological standpoint. The analytical data concerning these patients are set out in a series of tables, some of which are not commented upon since the facts are well known.

CLINICAL CONSIDERATIONS

Sex Incidence.—There are 216 males (77·9 per cent) and 61 females (22·0 per cent).

Age Incidence.—The majority of patients are in the age group 61–70 years. The distribution is shown in *Table I.*

Table. I.—CARCINOMA OF ŒSOPHAGUS: AGE INCIDENCE

YEARS	35–40	41–50	51–60	61–70	71–80	81–90
No. of cases	5	21	78	125	47	1
Percentage	1·8	7·6	28·1	45·1	17·0	0·4

Youngest, 35; Oldest, 82.

The majority of patients undergoing operation for this disease are old and present the general problems of surgery in the aged. The writer has operated upon a number with carcinoma of the œsophagus aged 70 years and over, and with careful pre- and post-operative care many of these patients stand the operation satisfactorily.

Duration of Symptoms (*Table II*).—An inquiry was made to determine the duration of symptoms before treatment was commenced, and the figures obtained show that in a large number of patients there is a long time lag. For example, in

Table II.—CARCINOMA OF ŒSOPHAGUS: DURATION OF SYMPTOMS BEFORE TREATMENT

DURATION IN MONTHS	Under 1	1–3	4–6	7–9	10–12	13–15	16–18	19–24
Number of cases	1	111	82	30	27	2	2	3
Percentage	0·3	40·0	29·6	10·8	9·7	0·7	0·7	1·0

In 1 case (0·3 per cent) the duration was stated to be many years.
In 18 cases (6·4 per cent) the duration was not stated.

25 per cent of patients the symptoms were present for periods longer than six months. Every endeavour must be made to bring the patient to the surgeon as soon as possible, and certainly within three months of the onset of the first symptom.

Early Symptoms.—A number of patients have been interrogated carefully regarding the first abnormality which was noticed, and it is apparent that the onset of the disease is very insidious. It is likely, moreover, that there is a latent period at its commencement, during which it is silent. A marked degree of proliferation of the growth into the œsophageal lumen must occur before dysphagia is produced and the same applies to stricture formation.

The patient is frequently unable to describe the initial symptoms with accuracy, and speaks of abnormal sensations beneath the sternum; an aching or burning pain, a queer feeling on swallowing, and perhaps a fleeting sensation of a food or liquid hold-up.

When the carcinoma is situated in the upper third of the œsophagus the first symptom may be dysphagia. For example, one patient described a sudden attack of dysphagia when drinking fluid and this was regurgitated with a quantity of phlegm. After this initial symptom, the dysphagia became progressively worse, especially for solids, and he was unable to eat meat.

If the carcinoma is situated in the middle third of the œsophagus the first symptom may be discomfort occurring after taking food. For example, one patient complained of this symptom nine months before admission. Four months later he experienced vague dysphagia which he thought was indigestion; he was able to swallow a few mouthfuls of food, when there seemed to be a stoppage at the level of the suprasternal notch. This disappeared and he was able to continue eating. Later he noticed the onset of vomiting after taking food, solids being worse than liquids, which he could keep down by sipping slowly. This symptomatology was noted in other patients.

With carcinoma in the lower third of the œsophagus the symptomatology is somewhat different and the first symptom is often the regurgitation of food. For example, a patient stated that one year before admission he had a sudden attack of regurgitation of food with excessive salivation. This occurred with solids and fluids.

The Presence of Metastases.—From the clinical standpoint it is important to form an opinion regarding the possible presence of metastases. This is an important method of differentiating the operable and inoperable cases. The view is taken that every patient with a carcinoma of the œsophagus should be regarded as a possible subject for surgical excision; the alternative to this is certain death. It is obvious, moreover, that every patient is not suitable for operation owing to the nature of the disease. With experience the surgeon can pick out the most suitable cases from the operative viewpoint, including those with a short history, partial dysphagia, minimal loss of weight and strength, and who are in good general condition. The patient with complete dysphagia, who is wasted, severely anæmic, and it may be cachectic, is often beyond radical surgical help. There are borderline cases, however, where an exploratory thoracotomy should be performed in order to determine operability.

Certain signs may be present which are indicative of advanced disease, apart from obvious ones such as enlarged liver, enlarged cervical lymph-nodes, pleural effusion, etc. These include a severe degree of anæmia, the presence of slight persistent fever, a clammy skin, and a marked loss of weight.

It is an important fact that in 42·5 per cent of patients where an autopsy was performed, there was no evidence of metastases. Even though no metastases are present, the growth in the œsophagus may be adherent to vital structures, including the descending thoracic aorta or the root of the lung, which precludes removal.

DURATION OF SURVIVAL WITH ALL METHODS OF TREATMENT, EXCLUDING SURGICAL EXCISION

In order to reach an estimate of the fate of patients with carcinoma of the œsophagus with all methods of treatment, excluding surgical excision, the after-history of each patient was investigated. The results are set out in *Table III*.

The figures set out in *Table III* demonstrate the killing nature of this disease; 67·1 per cent of the patients were dead within six months. Analyses were made in order to determine the results achieved with the chief methods of treatment available—namely, palliative surgery (gastrostomy), high-voltage X-irradiation, and radium therapy. The results are set out in *Tables IV–VI*.

Table III.—Carcinoma of Œsophagus: Duration of Survival with all Methods of Treatment, excluding Surgical Excision

Duration in Months	Under 1	1–3	4–6	7–9	10–12	13–24	25–36	37–48	84–96
Number Patients Dead	36	83	68*	26	13	23	6	1	2
Percentage	13.0	30·0	24·5	9.4	4·7	8·3	2·2	0·3	0·7

Number alive in 1947 1 (0·3 per cent)—4 years after treatment. Survival not stated in 16 (5·7 per cent). Number not traced 2 (0·7 per cent). * 1 patient killed through enemy action.

The figures show that after gastrostomy 53·2 per cent of patients died within 3 months and 75 per cent within 6 months.

Of patients treated by high-voltage irradiation, 30·6 per cent died within 3 months and 55 per cent within 6 months. This form of treatment has palliative value in that life is prolonged and for many patients dysphagia is temporarily relieved. More permanent benefit may result in isolated instances, as in the patient who survived for a period of 4 years.

The figures for radium therapy show that 37·2 per cent of patients died within 3 months of treatment and 76·7 per cent within 6 months.

Table. IV—Carcinoma of Œsophagus: Duration of Survival after Gastrostomy (92 *Cases*)

Duration in Months	Under 1	1–3	4–6	7–9	10–12	13–24	25–36	84
Number Patients Dead	12	37	20	12	6	3	1	1
Percentage	13·0	40·2	21·7	13·0	6·5	3·3	1·0	1·0

Table V.—Carcinoma of Œsophagus: Duration of Survival after High-voltage X-Irradiation (111 *Cases*)

Duration in Months	Under 1	1–3	4–6	7–9	10–12	13–24	25–36	37–48	84–96
Number Patients Dead	3	31	27	17	6	18	5	1	2
Percentage	2·7	27·9	24·3	15·3	5·5	16·2	4·5	0·9	1·8

1 patient is still alive 4 years after treatment.

Table VI.—Carcinoma of Œsophagus: Duration of Survival after Radium Therapy (43 *Cases*)

Duration in Months	Under 1	1–3	4–6	7–9	10–12	13–24	25–36
Number Patients Dead	6	10	17	7	—	2	1
Percentage	13·9	23·2	39·5	16·3	—	4·6	2·3

PATHOLOGICAL CONSIDERATIONS

Analyses have been made to determine certain points of pathological interest and the results are set out in *Tables VII, VIII.* No comment is made on the tables when well-known facts are illustrated.

Table VII.—Carcinoma of Œsophagus: Site Incidence

Site of Disease in Œsophagus	Number of Cases	Percentage
Post-cricoid	25	9·0
Upper third	31	11·1
Middle third	132	47·6
Lower third	81	29·2
Thoracic area	1	0·3
Middle and lower thirds	1	0·3
No sign of tumour (after X-irradiation)	2	0·7
Not stated	4	1·4
Total Number	277	

Table VIII.—Carcinoma of Œsophagus: Microscopical Examination

Microscopical Type of Neoplasm	Number of Cases	Percentage
Squamous-cell	168	60·6
Basal-cell	5	1·8
Gelatinous	1	0·3
Highly cellular, well-differentiated	4	1·4
Spindle-cell, epidermoid	1	0·3
Columnar-cell	8	2·8
Suggestive of carcinoma	1	0·3
No evidence of carcinoma	8	2·8
Specimen unsuitable	2	0·7
Section not taken	79	28·5
Total Number	277	

A study has been made to determine the position of relative frequency and distribution of metastases as shown by autopsy in 108 cases. Metastases were present in 62 cases (57·4 per cent), and absent in 46 cases (42·5 per cent). The distribution of metastases is shown in *Table IX.*

The metastatic picture shown in *Table IX* demonstrates that carcinoma of the œsophagus may produce widespread metastases and that nearly all organs of the body may be involved. The lymph-nodes are frequently affected, in the first instance those which lie in close proximity to the œsophagus, namely, at the bifurcation of the trachea and along the lateral aspect of the organ. Carcinoma in the middle third metastasizes to the former group in the first instance; and in the lower third to the second group. Attention is called to the frequency with which the lymph-nodes along the lesser curvature of the stomach and in the region of the cœliac axis artery are involved when the neoplasm is situated in the lower third. In all these cases it is essential to examine carefully these lymph-nodes and they must be excised when enlarged. In the upper third of the œsophagus metastases may occur in the cervical lymph-nodes. When advanced disease is present there is more widespread involvement of lymph-nodes.

Table IX.—Carcinoma of Œsophagus: The Metastatic Picture

Position of Metastases	*Number of Cases*	*Position of Metastases*	*Number of Cases*
Lymph-nodes (exact site not stated)	33	Pericardium	2
Supraclavicular nodes	2	Lung	16
Nodes at bifurcation of trachea	2	Bronchus	3
Œsophageal nodes	1	Pleura	2
Nodes around cœliac axis artery	6	Diaphragm	3
Inguinal nodes	1	Mediastinum	2
Liver	26	Femur right	1
Kidney	7	Elbow left	1
Adrenal	12	Tibia	1
Pancreas	7	Spine	1
Gall-bladder	1	Skull	3
Brain	2	Ribs	2
Thyroid	2	Sternum	3
Heart	3	Peritoneum	2
		Mesentery	2
		Transverse mesocolon	1
		Omentum	3

Metastases may be found in the lung or this structure is involved by direct extension. The secondary neoplasm often breaks down, becomes secondarily infected and a lung abscess is formed. The heart and pericardium may be involved by metastases or direct spread. The intra-abdominal viscera are frequently affected, especially the liver, where metastases occur frequently. When partial œsophago-gastrectomy is performed the liver must be palpated for the presence of metastases. There are several examples of osseous metastases in this series of cases, although the incidence is not high.

Local Spread.—In addition to local spread into the lung and pericardium, there are certain features of this aspect of the disease which merit attention.

Submucous spread in the wall of the œsophagus is of important clinical significance. There may be local invasion of the wall well beyond the obvious edge of the carcinoma and in the absence of ulceration this may be missed on œsophagoscopy. At operation for excision careful palpation must always be done in order to ensure that the line of section passes through normal tissue. When high-voltage X-irradiation is given in these cases and the fields of treatment are mapped out, allowance must be made for possible submucous spread of the disease beyond the borders of the tumour which is outlined radiologically. This fact may also account for failure after irradiation treatment in certain cases.

Spread of the neoplasm to the wall of the thoracic aorta will render it inoperable. In such cases well-marked stippling can be seen on the wall of the aorta. Differentiation is necessary between involvement of the peri-aortic cellular tissue and attachment to its wall. In the former cases the neoplasm can be stripped off with care. There may be involvement of the large blood-vessels in the hilum of the lung which renders the neoplasm inoperable. Attachment to the diaphragm need not necessarily preclude radical operation, for this may be excised.

Non-malignant Pathological Conditions.—On account of the age incidence of this disease it is not surprising to find that a large number of other lesions are present. It is important that the surgeon should be aware of the general level of health of these patients; therefore an investigation was made to determine the type and frequency of non-malignant lesions in this series of cases.

Pulmonary Lesions.—These are very common and include lobar pneumonia; bronchopneumonia; congestion; œdema; abscess leading to gangrene in some cases; bronchitis, emphysema, and pulmonary adhesions. Old healed tuberculosis may be present. Affections of the pleura are often seen, such as pleural effusion, empyema thoracis, pleurisy, pleural adhesions.

Cardiovascular Lesions.—This is a very common group of lesions in these patients. Various types of myocardial degeneration occur; hypertrophy and dilatation; and evidence of old endocarditis. The pericardium may be affected; these lesions include pericarditis, degeneration, and pericardial effusion. Atheroma of the arteries is common, including the aorta, coronary and carotid vessels. Coronary occlusion may be present.

Respiratory Passages.—These lesions include laryngitis and tracheitis, and purulent material may be present in the lumen. Œdema of the trachea and epiglottis are described.

Spleen.—Various affections of the spleen were noted and include perisplenitis, congestion, septic spleen, hæmorrhage into substance, and a cyst. In one case a metastatic abscess was present.

Liver.—Lesions affecting the liver include cirrhosis; angiomatous patches; toxic degeneration; congestion; perihepatitis; septic liver; nutmeg liver; adhesions between the liver and neighbouring viscera.

Stomach and Duodenum.—Affections of these organs include scarring; ulceration, both active and healed; benign tumours.

Kidneys.—Numerous lesions involving the kidneys were found, including congenital cystic disease; pyelitis; cloudy swelling; chronic nephritis; septic infection, and double ureter.

Other Lesions.—These are very common and include enlarged prostate; hydrocele of the tunica vaginalis; vesical diverticulum; ovarian cyst; fibroid uterus; thyroiditis; osteo-arthritis; Paget's disease of bone; abscess, congestion, and adenoma of adrenals; chronic cholecystitis; and ascites.

It is well to visualize this general health picture of patients with carcinoma of the œsophagus, especially when radical surgery is contemplated, so that careful pre- and post-operative treatment can be carried out with these factors in mind. These studies in the general pathology show that this group of patients is not characterized by a high standard of physical fitness.

SUMMARY

A clinico-pathological study is presented based on a series of 277 patients with carcinoma of the œsophagus. The early symptoms are described and correlated with the segment of the œsophagus which is affected. There is a considerable time interval between onset of symptoms and attendance at hospital for treatment—only 40 per cent of patients came within 3 months. The duration of survival has been determined for all methods of treatment (excluding radical excision) and after gastrostomy, high voltage X-irradiation and radium therapy. The metastatic picture in carcinoma of the œsophagus is presented. Attention is given to the presence of non-malignant pathological conditions in this group of patients; the results show that these are numerous and may be serious when radical operation is considered.

I wish to record my gratitude to Professor G. Grey Turner, who has done so much in this country to develop the surgery of the œsophagus, for his stimulating interest over a number of years. My colleagues at the Royal Cancer Hospital have allowed me to undertake these studies from the clinical and pathological aspects and I am grateful for their kindness to me. Professor D. W. Smithers kindly allowed me to quote the results obtained with high-voltage X-irradiation and Dr. M. Lederman those with radium treatment and I wish to thank them.

THE OPERATION OF PREFRONTAL LEUCOTOMY

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Moniz and Lima (1936) performed the first operations on the white matter of the frontal lobes of the brain for the relief of mental illness. At first they introduced alcohol through burr holes made in the vault of the skull, but later on by means of a wire loop they cut six cores on either side, each 1 cm. in diameter, at various levels in the frontal lobes.

Fiamberti (1937) approached the frontal lobes through the roof of the orbit, either injecting them with alcohol or incising the white matter, and this modification of the operation has been used by several others since that time.

Freeman and Watts (1937) planned the operation of lobotomy*; they opened the skull 3 cm. behind the orbital margin and 6 cm. above the zygoma on either side and cut as much white matter as was possible in the vertical plane immediately in front of the tips of the lateral ventricles, and they called this "the critical plane of section" (1942). The incision was made through a blind approach, but the coronal suture on the skull, the lateral ventricles, and the ridge of the sphenoid provided landmarks for the section, and it was checked by the injection of iodized oil and subsequent radiographs. Since that time the details of the operation appear to have been changed very little, though as a preliminary measure the brain is transfixed by introducing a guide between the two burr holes on either side of the skull. The method is described in detail in their book, and the subsequent refinements were fully demonstrated by their film which was exhibited at the Royal Society of Medicine in 1946. This technique has been largely followed by the surgeons in this country, and McKissock (1947), who has probably the greatest experience of the lobotomy operation here, described his method in Shorvon's paper (Shorvon and McKissock, 1947) as follows:—

"Burr-holes are made 3 cm. behind the lateral margin of the orbit and 5 cm. above the zygoma through 2-cm. skin incisions. The openings in the dura are large enough only to expose an area of cortex free from blood-vessels. A brain needle is inserted to a point 0·5 cm. short of the midline, the needle being angulated slightly forwards to avoid the anterior horn of the lateral ventricle. The needle, with stylet withdrawn, is then swept upwards and is, at the same time, introduced more deeply to an extent of 2–2·5 cm. to cut a line parallel with the falx, and finally outwards to divide as much white matter as possible It is then reintroduced in the original plane, swept downwards until just above the orbital plate, and finally swept outwards across the orbital plate to divide most of the white matter in this situation."

MacGregor (1941) invented an ingenious instrument (*Fig.* 101), used by Crumbie, by modifying a brain needle so that a rotating blade was inserted at one end and turned by a screw at the other. He also incorporated a sliding scale and a cannula from which the cerebrospinal fluid flowed if the ventricle was tapped in the operation. Since then MacGregor (1942) has produced a modification of his original instrument with a hollow needle and a spring which expands to a pear shape and which makes approximately the same-sized incision as his original leucotome. It has the advantage of pushing any vessels aside instead of cutting them as the white matter is divided. It is not, however, yet known to what extent the brain matter compressed by the expanding loop is capable of returning to its original position after withdrawal of the spring. The surface marking of Freeman and Watts was chosen for introducing the instruments, but only about a third of the total white matter in the plane was cut.

Fig. 101.—The MacGregor leucotome.

Knight (1943) used MacGregor's leucotome, but found it convenient to enlarge the incision by other means in certain cases.

Ross (1946) employs a rather different technique; though making his incision in a vertical plane he varies its anteroposterior position as much as 2·5 cm. in different cases according to the symptoms. He measures the distance back from the glabella instead of the orbital margin; most cuts are made anteriorly, but some posteriorly, to the critical plane of section. The appropriate position of the cut to be used in any particular patient was not, however, defined in detail in Frank's article (1946). Ström-Olsen, Last, Brody, and Knight (1943) also described the effects of incisions made posterior to the critical plane of section, as did Freeman and Watts (1942) in their book.

In an effort to identify the position of entry with accuracy and to control the bleeding, Lyerly (1939) made a larger window in the skull and performed the operation under direct vision; this practice has since been followed by some other surgeons in

* It has been suggested that the term 'lobotomy' should apply to these major sections and 'leucotomy' to the smaller operations where localized incisions are made.

America, but so far no paper upon its use has been published in this country.

Duff (1946) described a method which reverted to the original direction of approach from above instead of from the side of the skull, using his own special instrument; he believes it has the advantage of making a large incision of the white matter a safe procedure.

Freeman and Watts (1942) described the division of the lower quadrants of white matter in the usual plane, as did McKissock in 1943 and Smolik, Hoffstetter, and Busch in 1945. The writers (1943) described a horizontal leucotomy above the orbital surfaces and both they (1946, a) and Smolik, Hoffstetter, and Busch have claimed good results from the lower sections, though these do not yet appear to have been used very extensively by others.

Obrador (1947) described the first temporal leucotomy.

TYPES OF INCISION

The writers (1946, a) described seven different types of bilateral incision in the frontal lobes made by an approach through the temporal fossa, and it is with this work that the paper is particularly concerned.

Le Gros Clarke and Meyer kindly examined the brains of the five patients who died following operations made 3 cm. behind the orbit and 6 cm. above the zygoma. It was obvious that by using the same surface measurement in each case there were gross variations in the position of the incision. An approach was, therefore, sought whereby the constancy of the incision in the brain could be assured by seeing a definite landmark through a ½-in. trephine or burr hole. A spot was eventually chosen to make the opening in the skull 3 cm. behind the lateral orbital margin and 3 cm. above the zygoma, and the point of entry of the leucotome into the brain was determined by identifying the vessels in the Sylvian fissure just above the junction of the orbital and convex lateral surfaces of the cerebrum.

Having found a suitable point for the introduction of the leucotome, a method had to be devised to determine the direction and depth of insertion according to the fibres which were required to be cut and the size of the brain on which the operation was being done. The investigations were made on a number of fresh cadavers.

A point was marked on the scalp 3 cm. behind the lateral orbital margin and 3 cm. above the zygoma, and a trocar thrust through the scalp to make a mark on the bone. Through a small vertical incision the skull was cleaned, the mark identified, and the bone then drilled through to the brain. A sharpened spill dipped in a brightly coloured dye was then introduced through the hole just into the brain substance. On the opposite side of the skull the hair was parted and a line marked in the same vertical plane as that passing through the drill hole. This was divided into four equal lengths between the zygoma and vertex, or in other investigations into thirds or fifths, and differently coloured dyes were introduced, in the way already described, through holes at each of the marks drilled through

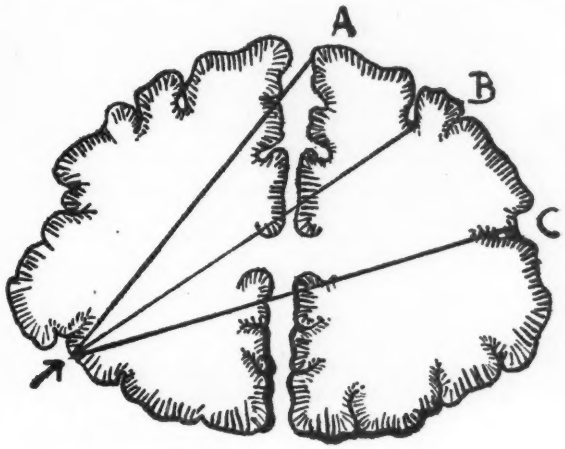


FIG. 102.—Tracing of section of the brain.

TYPE OF INCISION	DIRECTION OF LEUCOTOME	Distance inserted in cm. with a dura-to-dura measurement of: 100–105 mm.	105–110 mm.	110–115 mm.	115–120 mm.
Upper vertical	Towards a point 1 cm. outside the sagital suture on the opposite side of the skull in the vertical plane which passes through the point of insertion	5¼	5½	5¾	6
Middle vertical	Towards a point at the junction of the middle and upper thirds of a line between the zygoma and the sagital suture in the vertical plane of insertion	4¾	5	5¼	5½
Lower vertical (orbital)	Towards a point halfway between the zygoma and the sagital suture in the vertical plane of insertion	4¼	4½	4¾	
Horizontal	Towards a point on a horizontal level with the supra-orbital ridge and vertically above the outer margin of the orbit of the opposite side	4½	4¾	5	5¼

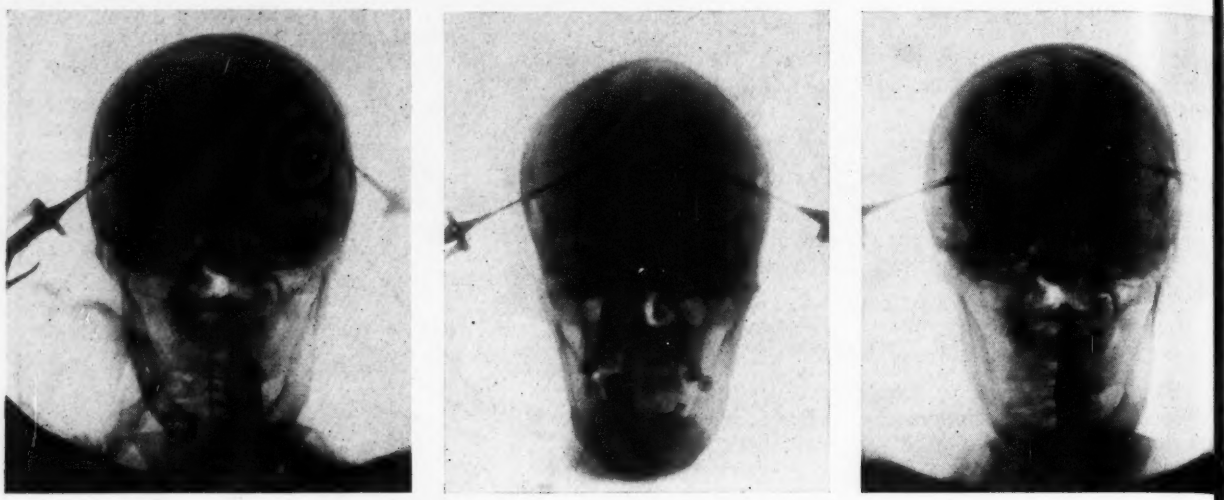
the skull in the direction of the opening in the opposite temporal fossa.

On the subsequent removal of the brains sections could be made through the vertical planes in which the various dye marks were seen. Next, whilst an assistant held a brain firmly in shape (freezing was unsatisfactory), an impression was taken of the freshly-cut surface by pressing transparent paper against it, the positions of the dye marks being drawn in the appropriate colours on the opposite or upper side of the paper. After the impression had dried these dyes were re-marked on the other or lower surface and the whole was then reproduced on to clean paper.

In this way full-sized tracings of the sections of the brains in which the operation incisions would have been made were obtained. It was then a simple matter to 'introduce' a leucotome on the paper, pointing it at one or other of the dye points on the opposite side of the brain, and to picture which fibres were cut when the instrument was introduced to any chosen depth. Moreover, in this way it was possible to see the maximum depth of incision which could be made to avoid the grey matter and the sulci with their vessels on the medial surface of the brain (*Fig.* 102). It was also possible to map out the planes of section of horizontal incisions by this means, but with greater difficulty, and ultimately the surface markings for horizontal incisions were made by measurements and sections on hardened brains in conjunction with skull examinations.

As a result of these investigations one or other of four different incisions have been selected for frequent use and at times a combination of two of them, in the treatment of appropriate cases. Three of the incisions are shown in the accompanying X-ray photographs which were made during operations (*Figs.* 103–105). The details of these incisions are shown in the Table; in each the leucotome is inserted by the temporal route.

Additionally, horizontal incisions have been made towards a point 1 cm. above the supra-orbital notch on a number of cases and in others towards a mark 1 cm. posterior to the usual horizontal surface marking (Reitman, 1946, a) and in two instances at an angle of 15° below the horizontal, but each of these methods has been abandoned. The only death as a result of the horizontal incisions was in a case when the leucotome was directed towards the point 1 cm. above the supra-orbital notch under a local anæsthetic. At the time when the vessel was injured the patient stated that something was being pulled out from behind her eye.



FIGS. 103–105.—Radiographs showing three of the incisions.

TECHNIQUE

The following are the technical details of the operation: Atropine $\frac{1}{100}$ gr. is injected, followed by intravenous pentothal, given in the ward if necessary. Intratracheal gas, oxygen, and ether has been found to be the anæsthetic of choice.

The head is completely shaved and prepared the day before operation. In a very few instances the patients have been so disturbed that an electrically-induced convulsion has been given to prepare them and to allow the anæsthetic to be administered. In one such case a small spider-like hæmorrhage was found on opening the dura.

The patient lies supine on a table so inclined that the head is considerably higher than the feet. The occiput rests in a ring of sorbo rubber. The tilting device on the table is used during the operation so that each side of the head in turn may be brought to a nearly horizontal position whilst being opened.

Incisions are made over points in either temporal fossa 3 cm. behind the outer margin of the orbit and 3 cm. above the zygoma. The incisions are slightly oblique so as to lie within the hair margin. The temporal muscles are split vertically and the fibres held apart with a self-retaining retractor. The bone is opened on either side by a burr hole which includes the upper and anterior part of the squamosal suture. The anterior divisions of the middle meningeal arteries are usually exposed lying in their osseous canals. A dura-to-dura measurement is made with a calliper of the pelvimeter type, as the reading, which may vary by over

20 per cent in different brains, is required for determining the depth of insertion of the leucotome.

The table is now levelled and the head settled firmly in the strictly supine position. The dura is opened by a cruciate incision and the vessels in the Sylvian fissure are identified; they are usually found just under cover of the bone at the postero-inferior margin of the burr hole. The pia arachnoid is punctured at a point where an imaginary line parallel to and 1 cm. in front of the vessels intersects a horizontal line just above the centre of the opening (*Fig.* 106). If necessary the bony aperture is

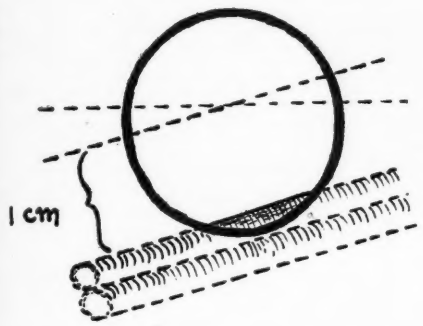


FIG. 106.—The site of puncture of the pia arachnoid.

enlarged upwards and forwards to expose this point, or if the vessels are not seen the opening must be extended backwards and downwards.

An appreciable error cannot be made in the vertical plane as the point of puncture is near enough to the orbital surface for this to be recognized if the approach should be too low and the Sylvian point is lost if the exposure is too high. The vessels in the Sylvian fissure give an accurate localization anteroposteriorly when taken in conjunction with the vertical measurements. The squamosal suture has also been found to be a very constant guide on the skull.

The scale on the MacGregor-Crumbie leucotome is now fixed for the required depth of incision, depending upon the width of the brain and the type of cut which is to be made, as has already been described. An assistant places the end of one probe on the point towards which the leucotome is to be directed and a second probe is held vertically on the glabella. Another assistant stands at the head of the table to see that the leucotome is inserted at right angles to the probe on the glabella. For the horizontal incisions the surgeon keeps the leucotome in the horizontal plane and the observer at the head of the table sees that the instrument is directed towards the chosen point; in this case the probe on the glabella is not required. The leucotome is inserted until its stop is in contact with the brain.

The leucotomes are constructed so that two or more rotations of the screw move the blade through 180°, the delicacy of control being such that the resistance afforded by a vessel can be felt. It is usual to rotate the leucotome blade back again to its original position before the instrument is withdrawn. Occasionally a vessel caught by the leucotome has been pulled out by the instrument and tied or coagulated before it has been released.

If the anterior horn of the lateral ventricle should be entered, the cerebrospinal fluid will flow from the side tube on the barrel of the leucotome. In this event the instrument must be reinserted to point more anteriorly.

When hæmostasis has been secured the dura is replaced without closure and the temporal muscle and scalp sutured in layers with interrupted stitches.

Post-operative restlessness must be avoided and soluble phenobarbitone (gr. 3) is therefore injected intramuscularly before the patient leaves the theatre and a chloral, bromide, and aspirin mixture is given rectally when the patient returns to the recovery room.

DISCUSSION

There has been much criticism of the operation, first for its blind approach and its inaccuracy, and, secondly, for the crude method of cutting the same mass of unselected fibres in an attempt to correct any form of mental illness. The operations which have been described go some way towards removing the objections on the first point and in regard to the second the succeeding observations appear to be of significance.

When the writers (1943) reviewed their first 50 cases operated upon in 1942 they mentioned the use of orbital leucotomy and the basis upon which the operation was designed.

As the result of twelve years of study on 105 cases of brain injury involving the frontal lobes, Kleist mapped out the cerebrum according to the supposed functions of its various parts, but generally he suggested that the convex lateral surfaces were associated with psychomotor and intellectual activities and the orbital surfaces could be considered the centre of emotional life. Injuries of the orbital region would thus give rise to a depressive displacement in the habitual feeling tone or more often the opposite displacement—euphoria, combined with "*Witzelsucht*" and puerility and sometimes moral insanity. Grunthal (1936) stated that emotional disturbances and abnormalities of behaviour, similar to those seen in psychopathy generally, occur after trauma to the basal surfaces of the frontal lobes and loss of initiative and intellectual deterioration after injury to the lateral convex surfaces.

Poetzl (1925), from a study of 2 patients with cysts, found that the akinetic syndrome was released from the convex lateral surfaces and the euphoria from the orbital surfaces.

Lemke (1936) in tumours and Guttmann (1930) in injuries found that changes in personality were particularly prone to appear in lesions near the orbital surface. Mayer-Gross (1938) drew attention to cases in which atrophy occurred on the orbital surface causing hilarious hyperactivity and lack of affective control.

Spatz (1937) made an important supposition that the inferior regions of the frontal and temporal lobes together form a functional unit which he called the basal cortex which has a special role in the regulation of emotional life.

Rylander (1939) showed that of 24 patients with parts of their orbital regions removed 18 became euphoric and 4 depressed, and that a rise in habitual mood towards euphoria occurred in pyknic types.

Earl Walker (1940) described the dorsomedial nucleus, pointing out the extensive representation of the orbital cortex in the medial portion of the nucleus, and suggested its function might be that of an integrating centre for somatic sensory impulses received from the lateral and ventral nuclei, and visceral stimuli relayed from the mesencephalon and hypothalamus, presenting them to the cortex of the prefrontal region in a more complex form.

Meyer and Beck (1945) demonstrated similar connexions of the dorsomedial nucleus to the orbital cortex in man, as had been shown by Walker to exist in the monkey. Bailey and Bremer (1938) and Bailey and Sweet (1940) found sensory cortical representations of the vagus by central stimulation of the nerve and also that respiration, blood-pressure, and gastric motility were affected by stimulation of the orbital cortex.

The writers (1943) stated that their upper incision in the operation of prefrontal leucotomy leads to improvement in cases with psychomotor activity rather than in the passive or emotional states, for which they suggested orbital leucotomy.

Shenkin and Rush (1943) showed that bilateral ablation of area 13 gave rise to hyperactivity, and Kennard (1944) described an increased production of adrenaline by this operation. Reitman (1946, b), from an analysis of the results of orbital leucotomy on the Netherne cases, believed that improvement from this operation arose by superimposing the triad of euphoria, hyperactivity, and extraversion on the initial symptoms.

McCulloch and Ward (1947) demonstrated a direct connexion between the frontal lobes and the hypothalamus by neuronography, and Reitman (1945), and Rinkel, Greenblatt, Coon, and Solomon (1947) have described autonomic changes following leucotomy. The latter writers believe that the changes they found are due to the liberation of the sympathetic and parasympathetic systems from the inhibition of prefrontal cortical centres.

It would appear that there is some anatomical, physiological, and clinical evidence in support of the selective incisions which have been employed, but it is hoped that still smaller and more localized sections may be made as the effects of the operation are better known (Dax and Radley-Smith, 1946, b). To this end a leucotome has recently been used which cuts an area the size of a sixpence. If comparable results to those with the larger incisions can be obtained by its use, much of the criticism of the operation will be obviated, whilst some additional knowledge of cerebral function may also be provided.

SUMMARY

1. The various types of operation used in the division of white matter of the frontal lobes of the brain for the relief of mental illness are described.

2. An account is given of the experimental procedure whereby a series of localized operations through an approach in the temporal fossa were devised.

3. Evidence is stated in favour of smaller and more selective leucotomy incisions varied according to the mental symptoms of the patients.

We are very grateful to Dr. F. Reitman, who has given us much assistance, and to Messrs. Down Bros., Ltd., for the loan of the leucotome block.

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RECURRENT EPITHELIOMA OF LIP

By PERCIVAL P. COLE, *O.B.E.*

CASE REPORT

Recurrent Epithelioma of Lip: Block Dissection of Anterior Triangle: Free Excision of Involved Area: and Reconstruction of Lip and Angle of Mouth at One Sitting.—

J. B., a stoker aged 28, was referred as a prisoner-of war from the Royal Herbert Hospital and came under my care at the Royal Cancer Hospital.

HISTORY.—In 1939 he cut the right side of his lower lip while shaving. An ulcer formed which did not heal and was treated by irradiation with radium in Belfast.

In October, 1941, two years after treatment, a recurrence was noted, and in March, 1942, he was admitted to the Royal Herbert Hospital. A biopsy taken there showed the growth to be an epithelioma. Wassermann and Kahn reactions were negative. He was referred to the Royal Cancer Hospital by the officer in charge of the surgical division and was admitted under my care on April 28.

CONDITION ON ADMISSION.—There was present a deep ulcer with raised edges measuring 1 cm. by 1·5 cm. just extending to the mucocutaneous junction and involving the lower lip and a corresponding area of the angle of the mouth. There was considerable surrounding induration but no fixation to bone. The right submaxillary gland was enlarged.

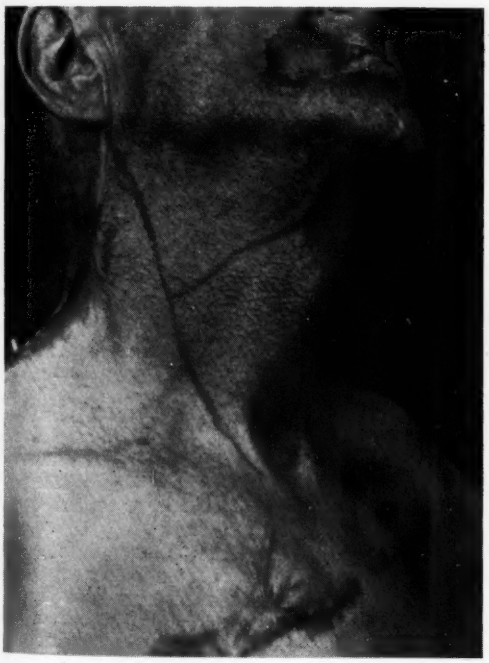

FIG. 107.—Usual incisions for approach to anterior triangle.

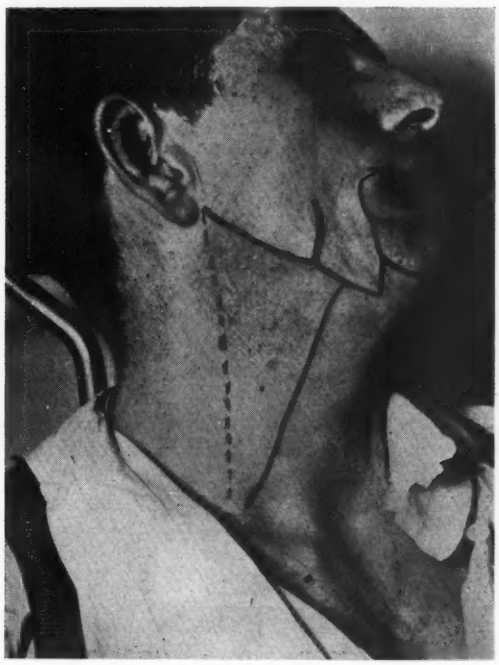

FIG. 108.—The incisions.

OPERATIVE TECHNIQUE.—The method of reconstruction adopted makes impracticable the approach to the anterior triangle by the usual incisions (*Fig.* 107). The somewhat irregular line (*Fig.* 108) tracing the incision along the lower border of the jaw at the base of the anterior triangle, divides the area of operation into two parts.

In the lower part is the anterior triangle with apex downwards, and free access to its contents is obtained by reflection of skin-flaps backwards and forwards from an incision joining its apex below to a point roughly midway along its base above (*Fig.* 108), the dotted line in which indicates the position of the anterior border of the sternomastoid.

Operation was performed jointly with Mr. Alan Hunt on May 2. The anæsthetic administered was gas and oxygen by intubation. The lower area was isolated from the face and mouth, and a block dissection was undertaken by way of the incisions referred to above (*Fig.* 108).

Exposure was considered adequate without removal of the sternomastoid and jugular vein, thus following in this particular case the technique of Butlin in preference to that of Crile. After clearance of the area the skin-flaps were united by suture, with dependent drainage at the lower angle of the wound. Following isolation of the area thus dealt with, the malignant ulcer with wide margin of surrounding tissue comprising the whole thickness of lip and adjoining cheek was excised. The flap for reconstruction was reflected and the process proceeded with as shown in *Figs.* 109–113. It is essential for success in this direction that mucous membrane be defined and sufficiently freed to be dealt with as a separate layer. Incision of this layer in the direction of the dotted line (*Fig.* 110) increases the length of the lower border of the mucosal flap and so permits it to reach to the midline if necessary and there to secure mucosal closure by suture to the corresponding layer of lip or to a corresponding flap from the other side if the whole lip has been removed. The knots in the suture line, *Fig.* 111, should be on the mucosal surface and are wrongly shown to make illustration more easy. The skin-flap is dealt with as in *Fig.* 112, and the gap thus resulting is easily closed without tension after the fashion shown bilaterally in *Fig.* 113. The method,

first indicated in outline by way of *The Lancet* (1920, **2**, 845) has been adopted as the standard type for reconstruction of the lower lip. As this case shows, the method is available where loss involves the angle of the mouth and encroaches upon the cheek to a limited extent. For obvious reasons, direct borrowing in such cases should be preferred to remote transfer of tissue, but the choice can only be made after a careful survey of the conditions present in the individual case. The condition ten days after operation is shown in *Fig.* 114. Due to an error in technique some infection occurred in the neck, but this had little effect on recovery and the patient was discharged from hospital on May 26, just over three weeks after the operation.

Fig. 109

Fig. 110

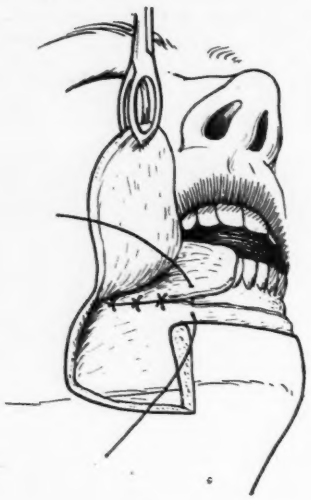

Fig. 111

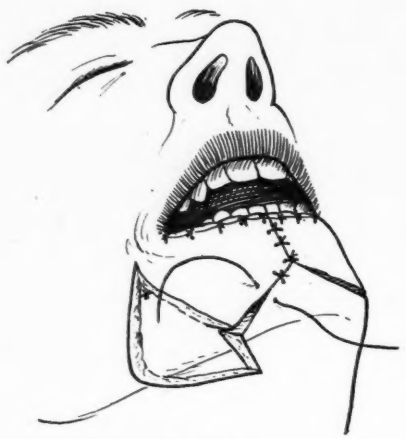

Fig. 112

FIGS. 109–113.—Steps in the reconstruction.

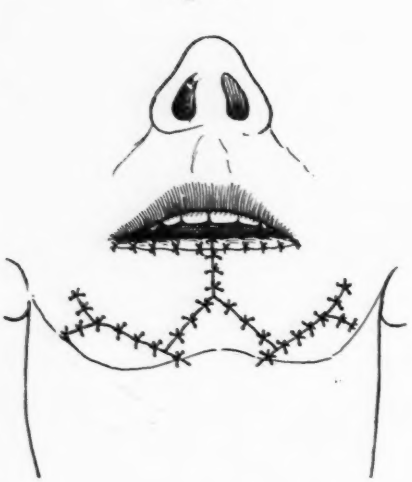

Fig. 113

Following operation subcutaneous nodules appeared, and the neck was heavily irradiated on the mistaken assumption that these were manifestations of incomplete removal. The scar in the neck developed keloid which was not resolved completely by the irradiation received. The patient left the country for his native home and was last seen on March 5, 1946. The photograph (*Fig.* 115) was taken in September, 1944, more than two years after operation, and the condition was practically unchanged when he was last seen on March 5, 1946, before his return to Austria. There was no sign of recurrence, the buccal sulcus was fully maintained in depth and capacity, the patient could whistle and had complete control over fluid, distending the cheeks as in the act of rinsing.

The keloid had diminished and the scar, though somewhat contracted, was pliable with no resultant functional disability.

POINTS OF INTEREST

1. The age of the patient. Epithelioma in this site is met with most frequently after the age of fifty. Occurrence at such an unusually early age has been in my experience associated with a breach of surface occasioned by a wound or burn. War wounds by bullet or shrapnel have been responsible factors in several such cases, and it is noteworthy that malignancy has supervened a very short time after their infliction. The same rapidity of onset is demonstrated in the present case.

2. Recurrence within two years after a dosage of irradiation that would have effected a cure in a large proportion of cases. Whatever view may be taken of the mode of treatment to be adopted for a primary lesion, recurrence after irradiation, computed to be adequate, is a clamant and unquestioned indication for surgical intervention. In this connexion the surgeon would do well to remember the pernicious behaviour of tissues in which permanent obliterative vascular changes have resulted from irradiation. To secure normal behaviour and to eliminate, as a preliminary to success, a factor the effect of which cannot be assessed or compensated, it is advisable to remove the whole area subjected to irradiation, as was possible in this case.

The possibility of failure and the resulting compulsory resort to surgery constitutes a strong argument in favour of methods which secure intensive localized irradiation in preference to those which employ irradiation on a more diffuse scale. It is indeed a question whether initially the surgical should not be preferred to the radiotherapeutic approach.

3. The completion in a combined operation of the threefold surgical requirements of removal of the focal lesion, featural and functional reconstruction, and clearance of the area into which growth may have extended by way of the regional lymphatics.

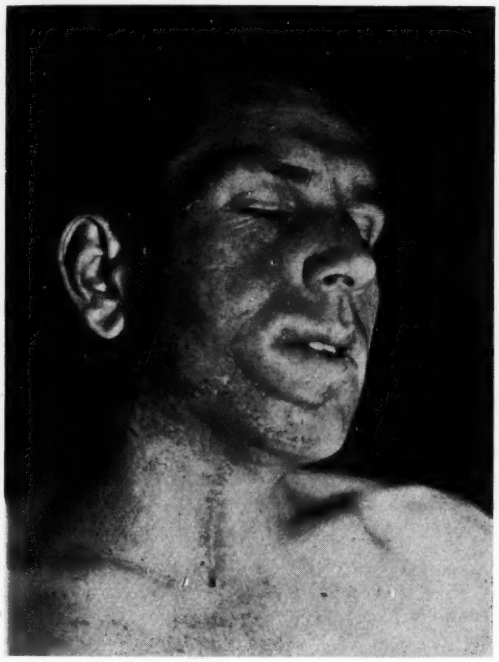

FIG. 114.—The condition ten days after operation.

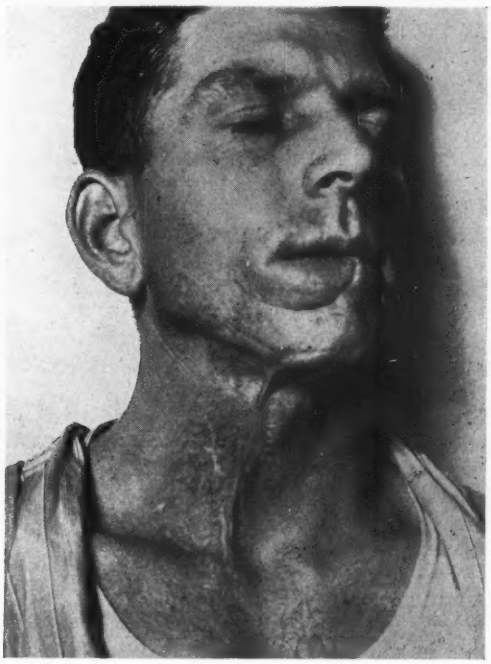

FIG. 115.—The patient two years after operation.

INTERSTITIAL-CELL TUMOURS OF THE TESTICLE

WITH A REPORT ON A CASE

By I. I. PRICE

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INTERSTITIAL-CELL tumours of the testicle are extremely rare, and, as far as I can discover, only 15 cases have so far been recorded in the literature.

Waldeyer in 1872 first described the interstitial-cell tumour, but in 1906 Chevassu reported the first authentic case and showed that Waldeyer's case was a seminoma.

Modern text-books of surgery fail completely to mention these tumours, and no case has been reported in the *British Journal of Surgery*. Yet

they constitute an extremely interesting section in the study of neoplasms of the testicle and their experimental induction in animals (Bonsor, 1940; Shimkin, Grady, and Andervont, 1942; Hooker and Pfeiffer, 1942).

The interstitial cells of the testicle or the cells of Leydig are epithelial cells of connective-tissue origin lying singly or in groups in the spaces between the seminiferous tubules.

Their numbers vary in different stages of the life cycle of the individual and they are also affected by the various diseases which may attack the individual.

In the fœtus these cells are very numerous while after birth they decrease in number and are replaced by fibrous tissue. As puberty approaches, they increase in number, while during adult life they decrease again, only to reappear as senility sets in and the seminiferous tubules atrophy.

Structure of the Interstitial Cells.—These cells are spheroidal or polyhedral in shape, with a small central or excentric nucleus containing chromatin and one or two nucleoli.

The cytoplasm contains highly refractile granules of neutral fat or lipoids and brownish coloured granules or lipofuchsin.

Hyperplasia of the Interstitial Cells.—This has been noticed in many conditions and has been experimentally produced. In some, hyperplasia has been so profuse as to simulate neoplasia. In the atrophic senile testes, Spongana found such a marked hyperplasia that in some cases the interstitial cells appeared to invade the walls of the seminiferous tubules.

Félizet and Brancha found interstitial cells actually in the walls of the seminiferous tubules in cases of undescended testicles, and they explain this on the assumption that the interstitial cells were developed from the connective tissue of the tubules.

Finnoti found a hyperplasia of the interstitial cells in all the 16 cases of undescended testicles he examined.

Lubarsch and Hansemann demonstrated hyperplasia of the interstitial cells in cases of chronic illness accompanied by cachexia, while Cordes noted the same phenomenon in cases dying of peritonitis and pneumonia.

Neoplasms of the Interstitial Cells.—Interstitial-cell tumours are common in dogs. On the experimental side of pathology, Kyrle irradiated the testicles of dogs sufficiently to induce minimal changes and found on section a hyperplasia of the interstitial cells.

Bonsor and Robson (1940), Hooker and Pfeiffer (1942), have induced interstitial-cell tumours in mice by the implantation of stilbœstrol-cholesterol pellets subcutaneously.

It is sometimes difficult to distinguish between hyperplasia and neoplasia of the interstitial cells, but to label the tumour as neoplastic it must be localized, and such localization must show an absence of seminiferous tubules. Cases of simple hyperplasia show a diffuse increase which is not confined to a single node and in some cases is so marked as practically to replace the testicular tissue and so suggest a neoplasm.

In most cases of tumour reported, there was a well-marked capsule, and in many the tumour constituted the major proportion of the testicle, the remains of which were compressed into a narrow band of degenerated seminiferous tubules.

When an interstitial-cell tumour arises before puberty, it is sometimes accompanied by signs of sexual and somatic precocity, as in the case of R. P. Rowlands and G. W. Nicholson (1929). Their case, a boy aged 9 years, showed signs of puberty from the age of 6. The testicle was five times larger than normal but orchidectomy caused no regression in bodily development.

Parkes Weber (1928) described a case of Sacchi, where removal of such a tumour in a boy of 9½ caused regression of sexual and somatic precocity.

Interstitial cell tumours are usually benign, except for a case of Masson and Sencert (1923) where metastases were found in the lungs, femur, and lymph-nodes.

A characteristic of these tumours is their brown mahogany colour, and the following case, occurring in an undescended testicle, enabled the diagnosis to be made immediately after operation.

CASE REPORT

A man, R. L., aged 53, reported with a left inguinal hernia. He had noticed the hernia for many years, but could give no definite time of its first appearance.

FIG. 116.—Painting of testicle. (*Natural size.*)

Examination showed a well-nourished man of full stature. The scrotum was empty. On the right side the testicle was not palpable in the inguinal canal or abdomen. On the left side a testicle smaller than normal was palpable in the inguinal canal and an indirect inguinal hernia was present.

Pubic hairs were present but there was an absence of hair on the chest. The breasts were of the male type. The voice was normal. He stated that he experienced sexual feelings, was capable of erection, but had no ejaculation. He was unmarried.

At operation, a left inguinal hernia was found and a radicle cure was performed combined with orchidectomy.

The testicle was smaller than normal and, on splicing it, the surfaces showed a brownish encapsulated tumour suggesting an interstitial-cell tumour.

THE SPECIMEN.—I am indebted to Dr. A. B. Bratton and Dr. C. C. Bryson, of the Archway Group Laboratory, for the following report: "The specimen consists of testicle, epididymis, and a portion of spermatic cord. The external appearance of the testicle is normal and it measures 4·5 × 3 × 2 cm.

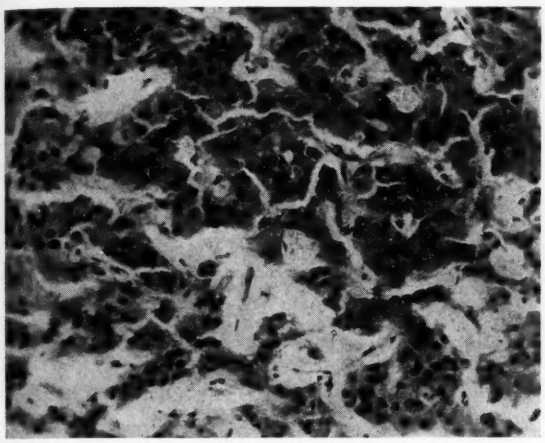

FIG. 117.—Microphotograph of section of tumour, showing large interstitial cells.

"The cut surface shows the usual finely striated yellowish testicular tissue; in the lower pole is a sharply circumscribed firm nodule (1·5 cm. diam.) which bulges from the surrounding tissue and appears as if it could easily be enucleated. The nodule is greyish-brown and is traversed by a few grey trabeculæ; it has a well-defined grey fibrous capsule. There are also a few grey nodules (up to 0·25 cm. diam.) in the surrounding testicular tissue.

"The epididymis and spermatic cord show no abnormality. (*Fig.* 116.)

"On microscopical examination, the nodule is composed of large polygonal cells. Their cytoplasm is eosinophil and occasionally contains fine light brown granules; many contain large or small droplets of fat, which stain orange with Scharlach R. The nuclei are well defined and vesicular and show an abundant fine chromatin network and a distinct nucleolus. These cells bear a close resemblance to the interstitial cells of the testis. They are for the most part arranged in large rounded solid masses in a dense collagenous stroma which forms wide curving trabeculæ continuous with a wide collagenous capsule. There are also small groups of cells in some of the fibrous trabeculæ and in the capsule. (*Fig.* 117.)

"The appearances are those of a localized adenomatous hyperplasia of the interstitial cells of the testis.

"The surrounding testicular tissue shows fibrosis of the stroma and atrophy of the seminiferous tubules, with fatty change in the germinal epithelium and absence of spermatogenesis. Throughout the testicular stroma there are numerous small groups of interstitial cells which have the same general appearance as those of the tumour. (*Fig.* 118.)

"A section including one of the small grey nodules seen naked-eye shows it to be an area of dense collagenous stroma containing an occasional interstitial cell and numerous tubules lined with hyperplastic testicular epithelium; it is apparently a localized area of adenomatous hyperplasia."

Further examination of the case a year later showed no suggestion of metastases.

The macroscopical and microscopical examination of this testicle leaves no doubt that this is a true case of an interstitial-cell tumour, for here we have a localized, encapsulated hyperplasia of the interstitial cells with a notable absence of seminiferous tubules.

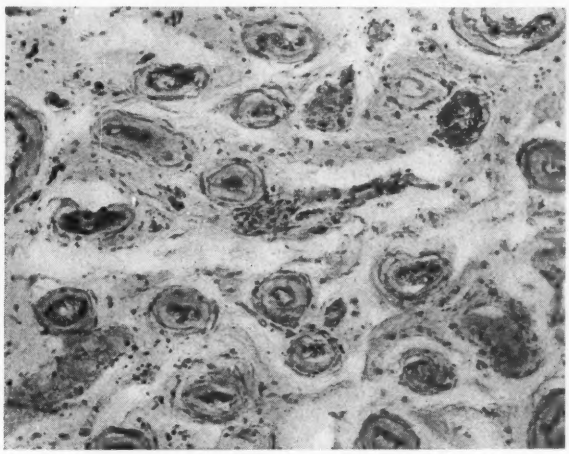

FIG. 118.—Microphotograph of section of testicle, showing atrophic tubules and some interstitial cells.

SUMMARY

1. The interstitial cells of the testicle or cells of Leydig are described.

2. The causes of hyperplasia of the cells are described.

3. A distinction is made between hyperplasia and neoplasia.

4. The literature on interstitial-cell tumours is reviewed and a case is reported.

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HISTOPATHOLOGY OF THE SEMISQUAMOUS EPITHELIAL LAYER IN THE SMALL INTESTINE

By FREDERIC DURAN-JORDA

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HAVING described elsewhere the histopathology of the semisquamous epithelial layer as seen in the stomach, colon, and appendix, we are now going to deal with the histology and some of the pathology of the layer as seen in the small intestine.

Compared with the stomach and colon, the small intestine gives only a minor proportion of surgical cases, and consequently, we have not been able to collect many fresh specimens. For this reason, our findings are not very extensive, but some of our observations are interesting and will help to give a new understanding of the histopathology of this organ, by taking into consideration the role played by the semisquamous covering layer in some pathological syndromes.

Coming from the gastric mucosa, the semisquamous epithelial layer covers without interruption the duodenum, jejunum, and ileum, and from there continues over the colon. In the normal small intestine, the layer appears to be very thin and fragile, and is destroyed very easily, not only by rough manipulation and the use of liquid fixatives, but also by autodigestion, if some parts of the mucosa remain in contact with a high concentration of enzymes, for our method of formalin vapour fixation actuates slowly.

FIG. 119.—Duodenum. Capillary included n the semisquamous epithelial layer.

FIG. 120.—Double mucous membrane structure as seen in the duodenum.

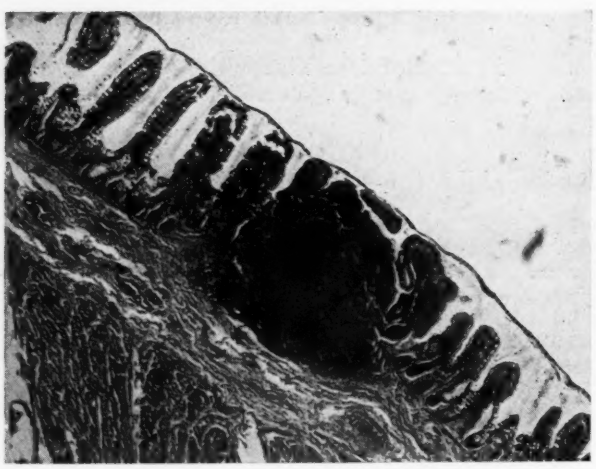

FIG. 121.—Ileum. The semisquamous epithelial layer following a parallel line to the axis of the intestine.

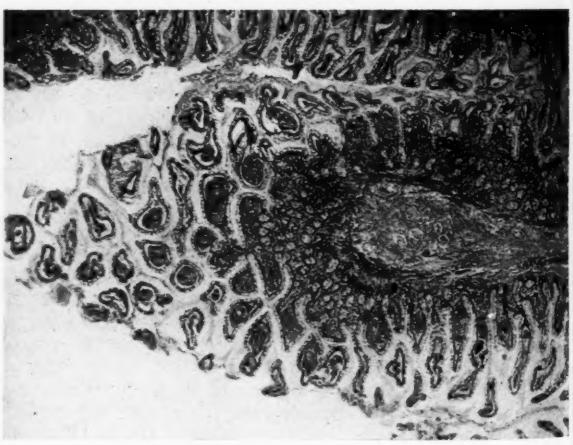

FIG. 122.—Ileum. Arabesque pattern. Notice that the semisquamous epithelial layer envelops every glandular island.

The layer is characterized by a quantity of diapedesic cells and it is interesting to notice that some of these diapedesic cells have a granulated cytoplasm which stains with eosin. The semisquamous epithelial layer also contains capillaries (*Fig.* 119), but in the intestine they are not so big as those found in the gastric mucosa. Like the layer found in the stomach, it does not take the

mucin stains, and after micro-incineration leaves a thin coating of ashes (Duran-Jorda, 1946). The contents of the small intestine are alkaline, and probably for this reason a thin semisquamous layer gives sufficient protection to the mucous membrane against the intestinal and pancreatic enzymes.

The normal small intestine studied by us, after formaline vapour fixation, has been observed to have three different structures :—

1. In some specimens of duodenum, there appears to be a double mucous membrane structure ; from the submucosa arise small villi which are protected by a semisquamous epithelial layer which

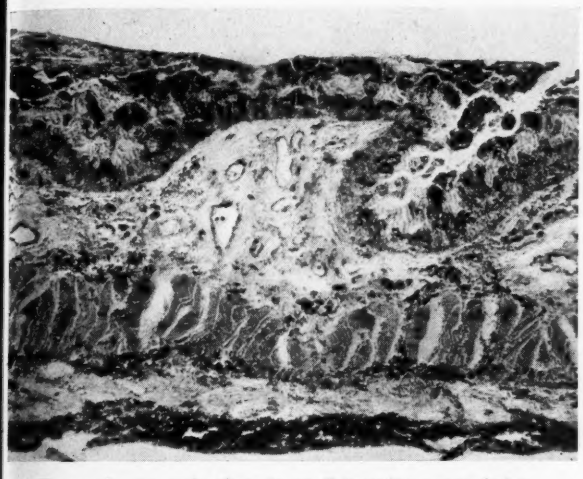

FIG. 123.—An early phase in small-intestine strangulation. Notice the hæmorrhage in the villi and the serosa.

runs along the top of the villi and contains diapedesic cells. Above these small villi there appear to be more villi, which are covered by another semisquamous epithelial layer (*Fig.* 120).

2. In the ileum portion of the intestine it can be seen that the structure is formed principally by long villi, covered by a layer which follows the line of the villi (Duran-Jorda, 1946).

3. In the duodenum and the last loop of the small intestine we have found short villi, with what appear to be islands of cells floating above them, all of which are covered but untouched by the semisquamous epithelial layer, which extends in a flat line parallel to the axis of the mucous membrane (*Fig.* 121). In some intestines these islands form a very decorative arabesque pattern, and surrounding every island there is a semisquamous epithelial layer (*Fig.* 122). This structure leaves empty spaces between the layer and the glands which *in vivo* are full of fluid produced by a double dialytic mechanism of secretion and absorption.

Strangulation.—Studying different resected specimens collected after small-intestine strangulations, we have been able to find two different phases. In the first phase there is a mass of hæmorrhagic infiltration in the villi and the serosa of the intestine, and the space between the layer and the villi becomes very thick with hæmorrhage and plasma extravasation. The action of the formalin vapour fixation at this stage produces a cast of the whole elements (*Fig.* 123). If the strangulation goes further, it involves the disappearance of the whole glandular structure of the villi, and the villi itself remains a mass of intratissular hæmorrhage—only in the base of the villi can still be found small remnants of the glandular structure (*Fig.* 124).

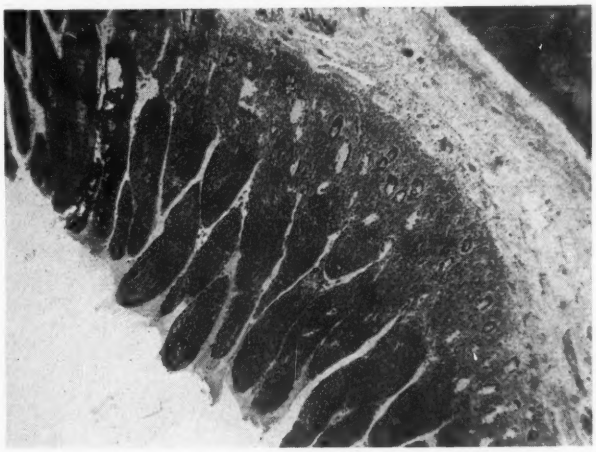

FIG. 124.—A later phase in small-intestine strangulation. The villi are a mass of intratissular hæmorrhage.

Ulcerative Colitis.—Ulcerative colitis as seen in the ileum, can, as far as we have observed, be divided into two different pathological pictures :—

1. In some places where the small intestine appears to be contracted, the semisquamous epithelial layer is thicker and produces a confluence of different parts of the mucosa (*Fig.* 125).

2. There are present cellular bridges, formed by inflammatory cells, which are between the

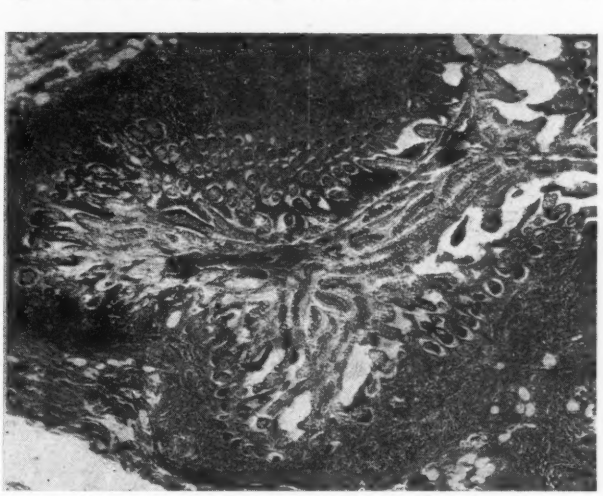

FIG. 125.—Ulcerative colitis as seen in the ileum.

semisquamous epithelial layer and the submucosa of a villus, similar to what we have described elsewhere as second-degree lesions in ulcerative colitis as seen in the colon.

Multiple Lymphomata.—In a case which was clinically suspicious of Crohn's disease, a part of the small intestine was removed, and we found macroscopically that the mucous membrane was hypertrophied, and contained multiple growths like lentils, which were not ulcerated. The section revealed that these growths were formed by a large

accumulation of lymphocytes in the submucosa, and there was atrophy of the mucous membrane itself. We diagnosed it as multiple lymphomata and not, as had been supposed clinically, a case of very early Crohn's disease. The semisquamous epithelial layer was normal throughout the intestine (*Fig.* 126).

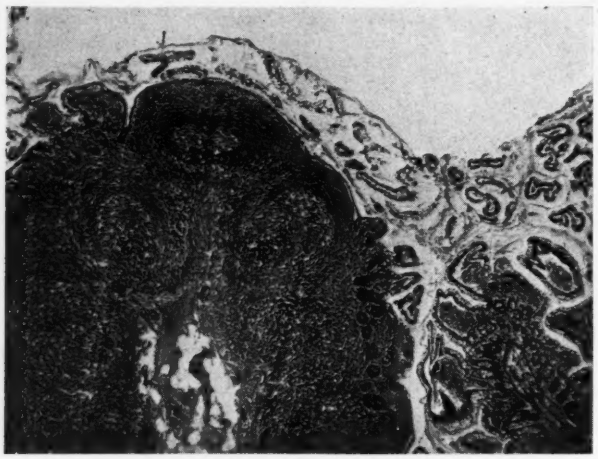

FIG. 126.—Multiple lymphomata of the small intestine. Notice the normal structure of the small intestine on the right of the photomicrograph.

Gaps in the Mucous Membrane.—If the portion containing the junction between the gastric mucous membrane and the duodenum is carefully prepared, it is very interesting to observe that at

FIG. 127.—A normal gap in the junction between the gastric mucosa and duodenum.

this point there is a big gap in the mucous membrane in some cases. Two facts supported the idea that the gaps were not pathological: One was the presence of the semisquamous epithelial layer covering the whole gap, and in some cases the layer came in contact with the Brünner glands; the other was the absence of any cellular infiltration. These gaps, as seen in the pyloric junction, were probably representative of an embryonic defect in the continuity of the mucous membrane of these two organs. (*Fig.* 127.)

In other parts of the small intestine smaller gaps could be seen occasionally. In some cases the gap was only two or three cells wide, and the non-cellular material of the submucosa seemed to come in contact with the space which exists between the villi and the semisquamous epithelial layer (*Fig.* 128). In other cases the gaps were occupied by a

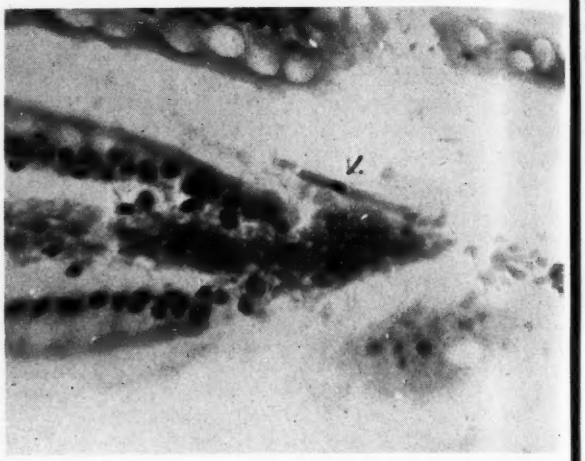

FIG. 128.—A small gap in the apex of a villus. Notice the capillary which has the diameter of one red blood-corpuscle (arrow).

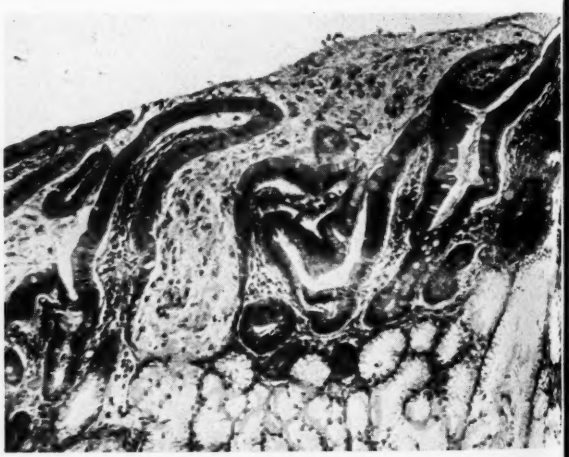

FIG. 129.—A gap in the mucous membrane which is filled by submucosa cells.

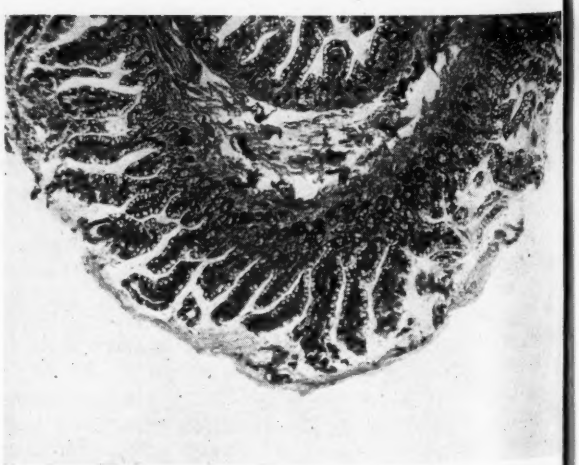

FIG. 130.—Meckel's diverticulum. Notice the normal aspect of the semisquamous epithelial layer.

bridge of cells which linked the semisquamous epithelial layer with the submucosa, and the cells which filled the gaps were characteristic of submucosa cells (*Fig.* 129). We were inclined to think that these gaps were a defect in the mucous membrane formation and were not characteristic of ulcer formation because similar gaps found in ulcerative colitis contained inflammatory cells.

Meckel's Diverticulum.—In specimens of Meckel's diverticulum we found that where the mucosa had a normal aspect there was a perfect semisquamous epithelial layer (*Fig.* 130).

SUMMARY

The author describes a very thin and friable semisquamous epithelial layer present in the small intestine, which has a diapedesic cellular content and contains very small capillaries. It does not stain with mucin stains and leaves a thin layer of ashes after micro-incineration.

He also describes the aspect of the layer in strangulation, ulcerative colitis, multiple lymphomata, and Meckel's diverticulum, and points out gaps found in the normal mucous membrane of the intestine which he considers have no pathological significance.

I am greatly indebted to the Surgeons of Ancoats Hospital for their kind co-operation with surgical specimens, and to Mr. J. B. Dean, B.Sc., for the photomicrographs.

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RADICAL MASTECTOMY

BY HEDLEY ATKINS

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SINCE the time of Halsted, radical mastectomy has been the treatment of choice in the early stages of carcinoma of the breast, and the results of Gordon-Taylor (1938), with surgery alone, set a standard which has never been seriously challenged. It should be our purpose, therefore, to make this procedure as safe and as curative as possible. In this connexion it may be urged that the functional, psychological, and cosmetic insults of a properly performed radical mastectomy are not appreciably greater than those following simple mastectomy; it is the idea of losing the breast that a woman deplores.

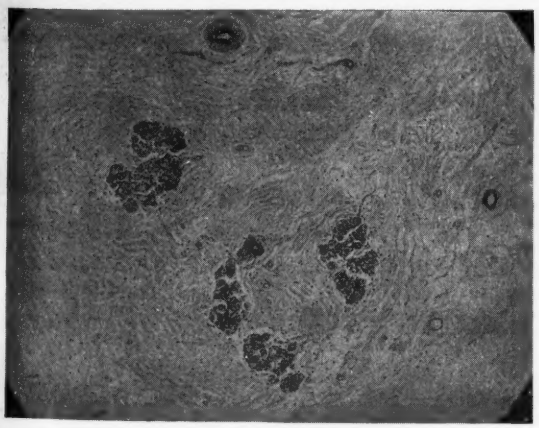

FIG. 131.—Section taken from a piece of breast tissue 3 in. away from an apparently localized carcinoma. Epithelium is hyperplastic and completely fills the lumen of the small ducts. (× 44.)

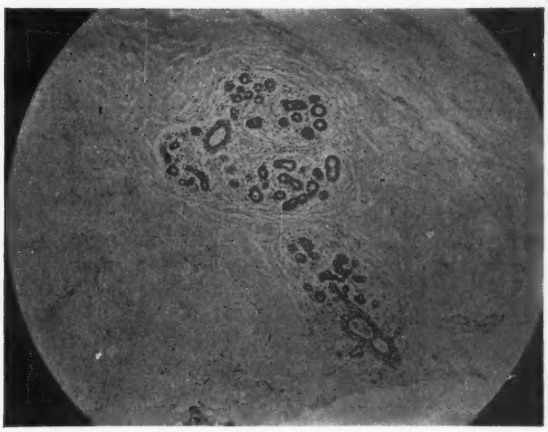

FIG. 132.—Section taken from tissue immediately under the skin, showing how areas of breast epithelium may lie in this region. (× 44.)

THE OBJECT OF THE OPERATION

In performing the operation the following criteria must be observed:—

1. Removal must be sufficiently radical. All skin within 2 in. of the palpable lump must be excised. The whole breast must be removed. Where frank carcinoma exists in one part of the breast, the epithelium of the ducts in other parts is often hyperplastic and in a premalignant state. To leave such epithelium behind is to court the possibility of recurrence (*Fig.* 131). This means that the skin over the breast tissue must be dissected on a very superficial plane from the incision outwards, and great care must be taken to ensure that no breast tissue is left adherent to the deep surfaces of the flaps (*Fig.* 132). There must be a thorough clearance of the lymph-nodes and fat from the axilla.

A wide area of deep fascia must be removed. Sampson Handley has for many years advocated this step, basing his advice upon the belief that there is a large plexus of lymphatics draining the breast and lying on the deep fascia. The existence of such a deep plexus has often been questioned

and even denied (Gray, 1936), but there is still some doubt about the matter, and in a recent paper (Oliver and Sugarbaker, 1947) this fascial pool is once more 'demonstrated'. Whilst there is this conflict of evidence it would seem wiser to assume the presence of deep lymphatics and to remove a wide area of fascia, particularly as this step adds nothing to the difficulties of the operation and, in fact, helps to define a plane where perforating branches of intercostal vessels may be secured.

2. The operation should be as bloodless as possible.

3. There must be practically no limitation of arm movement subsequently

4. Post-operative lymphatic œdema of the arm should be avoided.

Such desiderata are standard and traditional. During the last ten years I have made a study of the technique of radical mastectomy, and have introduced certain modifications which I believe implement these desiderata more fully.

The first modification is the infiltration of the skin with a solution of 1–500,000 adrenaline in normal saline both under the incision and deep to the skin-flaps. This infiltration serves two purposes: in the first place it renders the dissection of skin-flaps nearly bloodless, and in the second it defines precisely the plane of skin reflection so that this can be done with greater certainty at the correct level.

The second modification is designed to reduce blood-loss and loss of heat to a minimum. It consists substantially in leaving the reflection of the medial skin-flap to the last stage of the operation. Thus, the upper half of the lateral flap, and the upper few inches of the medial flap, are reflected first, so that the pectorals may be removed and the axilla dissected. Next, the rest of the lateral flap is reflected, and the mass of the breast and axillary tissues, supported first by the fascia covering serratus magnus and later by the pectoral muscles, is thrown medially by dissection deep to these latter structures. The pectorals are detached from their origins by approaching their deep surfaces, and dissection proceeds on a plane deep to the deep fascia until the sternum and xiphisternum are exposed. The whole mass is then turned back on to the hot towel which is covering the raw surface of the chest wall, and, finally, the now bloodless medial flap of skin is reflected so that the parts to be removed are released. This method allows the time-consuming axillary dissection to be carried out before large areas of raw surface are exposed. It ensures that the perforating branches of the intercostals are visualized as they traverse the relatively translucent areolar tissue deep to the fascia covering serratus magnus and deep to the pectoral muscles, so that their solitary parent trunks can mostly be secured before they are cut. It obviates the necessity of grabbing the many superficial branches of these vessels as they traverse the opaque fat deep to the medial skin-flap, and later cutting the parent vessels when the pectorals are reflected; and finally by a combination of these effects it provides the patient with an immediate post-operative period free from the anxieties of shock, and a convalescence which is correspondingly rapid.

Stanford Cade (1940) has emphasized the high incidence of recurrence in the skin following the standard methods of radical mastectomy. It is hoped that the wide area of skin removed and the precise clearing of the flaps which the injection method allows, will serve to reduce the incidence of such recurrences.

THE OPERATION

As many of the details of the operation to be described follow precisely the standard Halsted technique, it will not be necessary to discuss each step, but only to indicate where these have been modified, and to mention certain anatomical considerations which are insufficiently stressed in the standard accounts.

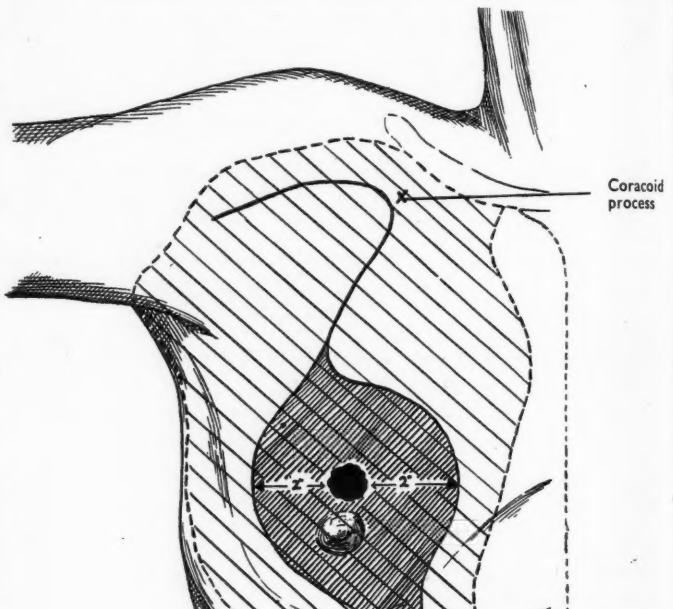

FIG. 133.—The incision is indicated by a continuous line, and the outer dotted line shows the limits of flap reflection. The light shading represents the area infiltrated subcutaneously with adrenaline solution.

1. After the skin has been sterilized and the arm fixed in the abducted position, the lump is precisely defined and a circle scratched on the skin 2 in. distant from the edge of the lump. This circle must not be encroached upon. The rest of the incision is then marked out by scratching with the scalpel blade. It runs from the coracoid process above, downwards to embrace the circle first marked out; then downwards and medially to a point an inch below the xiphisternum. A small extension at the upper end of the incision is carried laterally from the coracoid process down the arm for 2 in. parallel to the lower border of the

deltoid muscle, giving a semicircular flap hinged laterally and nourished by branches of the subscapular artery.

The line of the incision is then infiltrated with a solution of 1–500,000 adrenaline in normal saline, extending out from the line of the incision, medially to the limits of breast tissue, above to clavicular level, laterally to the anterior border of the latissimus dorsi, and below to the epigastrium (*Fig.* 133). This infiltration is conveniently performed with an automatic-filling syringe and 10 oz. of saline solution are used. A further infiltration is sometimes made into the axilla, so that tissues can readily be dissected subsequently, but if this step is adopted a simple syringe must be employed so that the plunger may be withdrawn before any fluid is injected. This procedure, as indicated above, defines the level of dissection and acts as a hæmostatic. As the fluid is injected into the plane of future dissection there is no danger of disseminating cancer cells, even if, as is extremely doubtful, dissemination were possible by such a procedure.

2. The upper part of the incision down to the point of divergence to encircle the neoplasm is now made, taking care that the knife cuts through the skin only, wherever the incision overlies breast tissue. The upper part of the lateral flap is reflected to expose the lower border of the pectoralis major muscle, the deep fascia forming the base of the axillary pyramid, and the upper end of the anterior border of the latissimus dorsi. The upper few inches of the medial flap are reflected in order to expose more of the pectoralis major muscle, this part of the medial flap being fortunately free from large perforating blood-vessels.

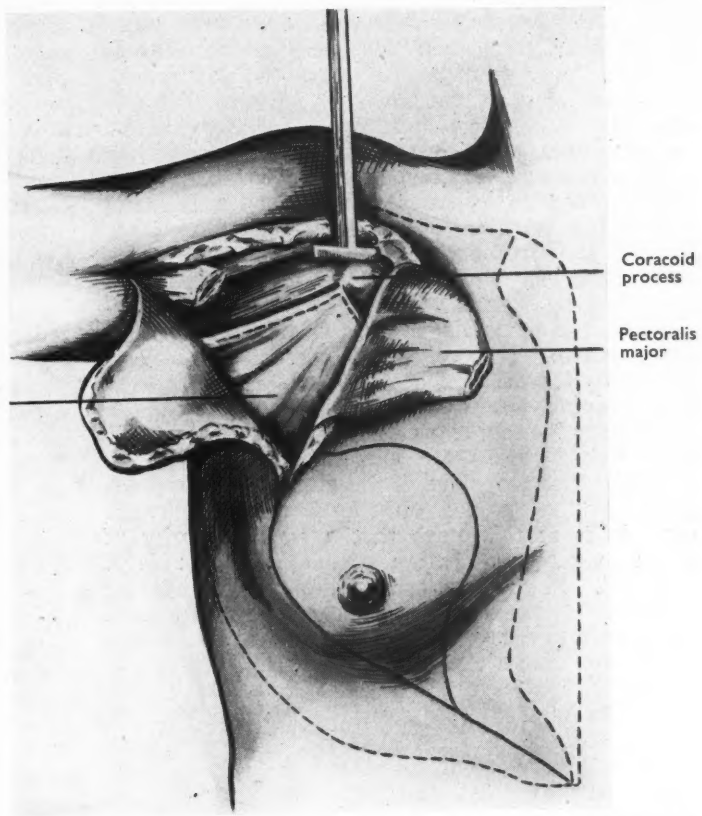

FIG. 134.—The brachiopectoral fascia is incised along the dotted line. This enables a clean dissection to be made of the lower border of the pectoralis minor and the medial border of the coracobrachialis.

3. In most instances only the sternal head of the pectoralis major is removed, so that the gap between this head and the clavicular head is defined and split to expose the fascia on its deep aspect. In view of the U-shaped insertion of this muscle, the guide which will define the deep surface of the tendon when the insertion is cut through must be passed from below (from the base of the U), otherwise it will enter between the two limbs. The lower border is then clearly defined, and a guide passed deep to the pectoralis major to emerge in the gap made between the sternal and clavicular heads. The insertion of this muscle is detached as near as possible to the bone.

4. Although the costocoracoid membrane is dignified by a name, it is usually a far less definite structure than is that part of the deep fascia which bridges the angle between the lower border of the pectoralis minor and the medial border of the coracobrachialis, a fascial layer which passes downwards and laterally to blend with the deep fascia over the serratus magnus (*Fig.* 134). This fascia should now be cut with a knife along the medial border of the coracobrachialis, and along the lower border of the pectoralis minor, the upper ends of the incisions meeting at the coracoid process. This fascia, which I have called the brachiopectoral fascia, is now reflected downwards from the apex by a combination of sharp dissection and gentle brushing with the gauze-covered finger. Its removal reveals the axillary contents in their lateral parts, and clears the lower border of the pectoralis minor. The upper part of the brachiopectoral fascia—namely, the costocoracoid membrane—is now exposed by retracting the pectoralis major medially, and this fascia is incised along the upper border of the pectoralis minor. A protecting guide can now be passed deep to the pectoralis minor and close to its insertion where the latter is cut and reflected medially.

5. The cutting of the nerve- and blood-supply to the pectoral muscles allows these muscles to be hinged more freely, and the whole contents of the axilla are exposed. The dissection of these contents proceeds according to standard technique, and all fat and glands are removed cleanly. The thoracodorsal nerve to latissimus dorsi is usually preserved, and the long thoracic nerve nearly always so. I have made a habit of passing a finger into the apex of the axilla in front of the neurovascular bundle, and brushing down the fat deep to this bundle and removing it, but as in 34 consecutive

cases in which there were glandular deposits in the axilla, this fat has proved on microscopical examination to be free from invasion, and as the procedure is attended by some oozing from small veins difficult of access, it is doubtful whether this manœuvre is worth while.

6. Having cleared the axilla, a gauze sponge wrung out in warm saline at 110° F. is inserted into the cavity and the rest of the lateral flap is completed, cutting superficially and as far as the anterior border of the latissimus dorsi. The plane of the incision is then deepened, and the fascia covering the serratus magnus is exposed and incised, the lower end of the incision curving forwards across the wall of the chest and over the abdominal muscles to a point just distal to the tip of the xiphisternum. Dissection with the blade of a scalpel readily separates this deep fascia, and perforating branches of the intercostal vessels are secured and cut as they emerge through the intercostal spaces. The plane becomes more and more clearly defined as the level of the deep surfaces of the pectoralis minor and pectoralis major are reached. The origin of pectoralis minor is detached, leaving a felting of muscle wherein a few bleeding points can readily be secured. As the deep surface of the pectoralis major is approached, many large vessels are encountered, and as the origin is detached, some of these, hidden in muscle-fibre, can be secured only after they have been cut. It is at this stage that a small quantity of blood may be lost before adequate hæmostasis is obtained. Having detached the origin of the pectoralis major muscle, dissection proceeds on a plane deep to the deep fascia until the bony sternum is exposed across the midline. During the course of the reflection of this large, muscular, fatty, cutaneous mass, a raw surface is exposed on the lateral chest wall, and this is kept continually covered with a small towel wrung out in saline solution at 110° F., and from time to time the perforating vessels, secured by hæmostats, are coagulated with the diathermy electrode.

7. The large flap containing the breast, muscle, and axillary contents is then turned back on to the hot towel. The now bloodless medial cutaneous flap is reflected, at first very superficially, but as the dissection proceeds beyond the confines of the breast, more deeply to meet the deep dissection just beyond the midline. This frees the parts to be removed, which are wrapped in the towel on which they lie and handed to the assistant. A few vessels may require coagulation, but usually the skin-flaps can be immediately sutured, with the insertion of such drains as the nature of the cavity may seem to demand.

8. Skin suturing is best performed with interrupted sutures, so that there is as little strangulation of the skin edge as possible. With the wide skin excision advised, complete closure is impossible in about half the cases, and in these the resulting raw area is immediately grafted with 'postage-stamp grafts' of split skin taken from the thigh. Tulle gras and gauze sponges soaked in flavine are placed over the grafted area.

9. The dead space in the axilla must now be obliterated, and this is performed by means of a cotton-wool snowball (4 in. in diameter when compressed) thrust firmly up into the axilla. A firm 'many-tail' bandage holds this snowball in position and allows the arm to be nursed in 70° of abduction without danger of a serous collection in the axilla. It is not infrequently the organization of such serous collection, with consequent fibrosis, which is responsible for the strangulation of such lymphatics draining the arm as are left behind, and the development of post-operative lymphatic œdema.

10. Drainage tubes are partly withdrawn after forty-eight hours, and completely withdrawn on the fourth day. Grafted areas are dressed on the seventh day when the stitches are removed.

Convalescence is usually uneventful. Shock is reduced to a minimum, and the patient may get out of bed within a week. Arm movements are encouraged, within the limits that the firm bandage allows, from the second post-operative day, so that at the end of a week the patient should be able to place the hand on the top of the head, and within ten days the opposite ear can be touched with the head erect.

SUMMARY

1. Radical mastectomy is the treatment of choice in early carcinoma of the breast.

2. The objects of radical mastectomy are defined.

3. Recurrences after the standard methods have been commonly encountered in the skin. These skin recurrences are due to a niggardly removal of skin, and to leaving breast debris behind in the deep surfaces of the flaps.

4. A method of radical mastectomy is described using the infiltration of a weak solution of adrenaline deep to the flaps. This infiltration controls hæmorrhage and defines precisely the layer to be cut in reflecting the flaps. Hæmorrhage is further controlled by dissecting the axilla first, securing the trunks of the intercostal vessels as they emerge, and finally reflecting the medial flap.

5. Raw areas are covered by a 'split-skin' graft, and dead space is obliterated by means of the cotton-wool 'snowball'.

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DEFECT IN THE BROAD LIGAMENT AND ITS ASSOCIATION WITH INTESTINAL STRANGULATION

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THE occurrence of a defect in the broad ligament is of some rarity, for reports of, or references to, only 45 cases have been found in the literature. Of these, 34 have been traced in the original articles; 7 case reports have not been available for consultation; details of 4 cases are obtained from the paper of Füssel (1937).

Seventeen of the 45 cases have followed the performance of a uterine suspension operation of the Webster-Baldy type; in this procedure, holes are made through the broad ligaments below and medial to the utero-ovarian ligaments, and through these the round ligaments are drawn before being sutured to the posterior surface of the uterus and to each other. In the remainder, as in the case described here, there has been no known direct interference with the broad ligament. It is a striking fact that in all but 5 of these patients with broad ligament defects, attention was first drawn to the condition by the occurrence of intestinal strangulation. In one of the exceptions symptoms were produced by the passage of the ovary through the aperture (Masson and Atkinson, 1934).

When the intestine, usually the lower ileum, has been found strangulated in an aperture in the broad ligament, the condition has frequently been described as one of 'strangulated internal hernia'. In fact, in half the cases the defect has involved both layers of the broad ligament, and the gut has passed right through it, the state of affairs being comparable to that of strangulation by a band. At other times the defect has been in one layer of the ligament only, and the intestinal loops have become incarcerated between the layers. Fagge (1917) (2 cases), Masson and Atkinson (1934) (in whose case the ovary had migrated into the defect), Dorman (1928), and Hermann (1925), state that in their patients a peritoneal sac was present, derived from the defective layer of the broad ligament. Both Janes (1929) and Hunt (1934), however, emphasize that the peritoneum covering the incarcerated bowel in the cases observed by them was derived from the intact layer of the broad ligament, and this was so in Finlaison's case and in that reported here. A sac has been demonstrated only in five patients: the involved bowel more commonly remains in the general peritoneal cavity, and may as often enter the parametrial tissues.

CASE REPORT

Mrs. R. S., aged 43, a multipara with a history of four normal labours, her eldest child being 21 and the youngest 6, was admitted to Hammersmith Hospital in emergency, suffering from abdominal pain of thirty-hours' and vomiting of twenty-four-hours' duration.

The abdominal pain began at 10 a.m. on the day before admission, suddenly and without cause. It was very severe and colicky in nature: during the short remissions of colic she was aware of a continuous pain which was much less severe. The pain at first was para-umbilical, but after two to three hours it shifted to the right iliac fossa, and later still returned to the centre of the abdomen. It did not radiate to the back or the groin. Some six hours after the onset of the pain, the patient vomited, with slight relief. Thereafter only fluids were taken, and these were returned within ten minutes. A normal stool was passed about five hours after the pain began. There were no urinary symptoms. The last menstrual period had ended a fortnight before admission, and had been quite normal. There was no vaginal discharge, nor were there any other gynæcological symptoms.

ON EXAMINATION.—The patient was a thin woman, pale and anxious, and obviously in considerable pain. The temperature was 99·8°, pulse 80 per minute, and respiration-rate 16 per minute. The face was rather drawn and the lips dry, but the tongue was moist and clean. The breath was not unpleasant. No abnormality was detected in the chest. The systolic blood-pressure was 135 mm., the diastolic 90 mm. Hg.

All quadrants of the abdomen moved equally with respiration; there was no distension or visible peristalsis. The muscles of the right lower quadrant were resistant to palpation. There was tenderness and rebound tenderness in the left iliac fossa, and very great tenderness in the right iliac fossa, maximal at McBurney's point. Pressure on the left side did not cause pain on the right (Rovsing). Movements of the hip in any direction evoked no pain. There was hyperalgesia to pin-prick in the triangle bounded by the highest point of the right iliac crest, the right pubic spine, and the umbilicus (Sherren's triangle). Tenderness and rigidity were absent in the upper abdomen. On auscultation, the bowel sounds were slightly increased in volume and frequency. Rectal examination revealed tenderness in all directions: blood was not withdrawn on the examining finger. Vaginal examination proved the uterus to be retroverted and normal in size. No abnormal swelling was felt. There was generalized tenderness on pressure in the posterior fornix. Because of the copious vomiting it was felt that there was more reason to suspect an intestinal obstruction than an acute obstructive appendicitis, which seemed, after obstruction, the likeliest diagnosis. Accordingly a paramedian rather than a more lateral approach to the abdomen was chosen.

OPERATION.—Anæsthesia was induced by nitrous oxide and oxygen and maintained by cyclopropane. A lower right paramedian incision was made. As soon as the peritoneum was opened there was a gush of clear, yellowish fluid, which was removed by suction. A hand was passed towards the pelvis, and as it reached the neighbourhood of the uterus a small aperture with firm edges was felt. This was taken to be an internal hernial orifice of doubtful site. The table was tilted into the Trendelenburg position, packs were inserted, and it was possible to demonstrate dilated small bowel running up to the orifice which had been felt and which was now seen to be situated in the right broad ligament (*Fig.* 135). A finger was inserted into it and its margins were stretched. A gush of blood-stained fluid followed this, and subsequently some 6 in. of strangulated lower ileum were withdrawn. The loop of intestine was blackened and the peritoneal coat peeled off at the gentlest touch. The mesentery was pulseless and stained by hæmorrhage. There was no evidence of improvement after wrapping in warm packs for five minutes. Accordingly the loop was resected and an end-to-end repair of the ileum was carried out with two layers of suture.

The defect was now examined (*Fig.* 136). Before dilatation it had been just large enough to admit the tip of the little finger. Its edges were firm, white, puckered, and well-defined. It was situated between the right round ligament and the Fallopian tube, about an inch from the margin of the uterus. There was no hernial sac. The gut entered from above and in front to lie between the layers of the broad ligament.

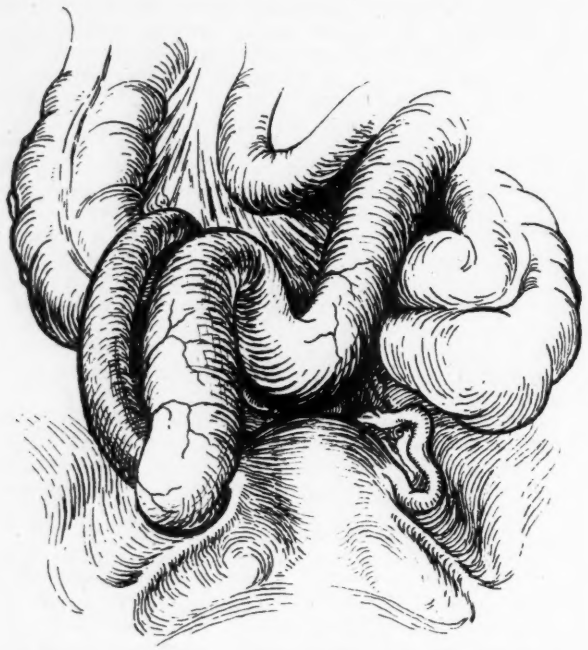

FIG. 135.—Condition as seen at operation. Dilated small intestine running up to defect, collapsed bowel leaving it.

The aperture was repaired by a running catgut suture and the abdomen was closed. Convalescence was uneventful and the patient left hospital twenty-one days after operation.

DISCUSSION

The first instance of intestinal strangulation due to a broad ligament defect was reported by Quain in 1861, as an autopsy finding. Since Quain's description, cases have been reported singly and in pairs by many observers. It is not proposed to include here the apertures which have been found in patients who have undergone the Webster-Baldy operation. The association of these with the operation, and the technical errors which may be responsible for their occurrence, have been very fully and clearly discussed by American writers (Arnold, 1938 ; Parkes and Karabin, 1939 ; Pulrang, 1944 : Pemberton and Sager, 1929).

Aetiology.—The case reports of patients who had not previously undergone operation have been studied in detail in relation to the following points :—

1. *Previous Pregnancy.*—All the patients except four have been parous women, mostly with a history of normal labours, though one had both children delivered by a Cæsarean section. Symptoms of intestinal involvement have never arisen during pregnancy, though in one patient they appeared very shortly after delivery. (Dunn's (1926) patient developed abdominal pain following a fall, and she tolerated it for fifteen years before acute intestinal strangulation made operation necessary. It is interesting that this pain was absent only during her numerous pregnancies.)

2. *Age.*—Most of the patients have been in the middle years of life. The youngest was 18, the oldest 79, but the average age is 44.

3. *Previous Pelvic Inflammation.*—Only 2 patients, Quain's original case and that of Finlaison (1946), had any history or signs of past pelvic inflammation.

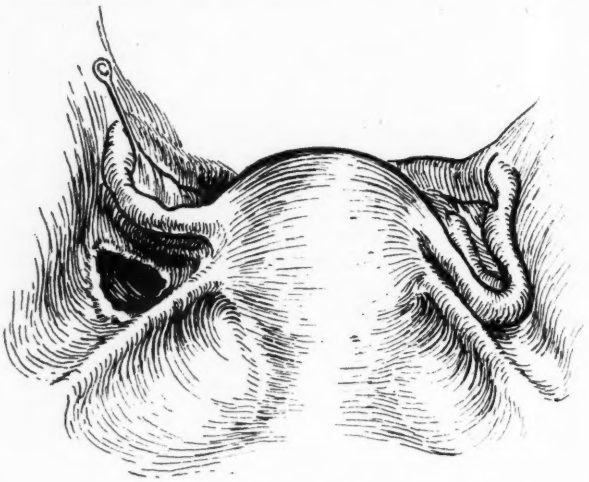

FIG. 136.—The defect displayed after withdrawal of strangulated bowel.

4. *The Position of the Uterus.*—Has been variable and not always definitely described.

5. *The Nature of the Defect.*—In 11 of 22 cases in which the relation to the layers of the broad ligament has been mentioned, the defect involved both layers. In the other half, one layer only was affected, but in varying ways. Pidcock (1924) and Goode and Newbern (1944) describe an opening in the ' mesoligamentum teres ', by which the round

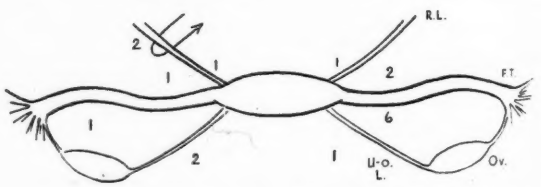

FIG. 137.—Diagram to show the incidence of defects at various sites in the broad ligament. Numbers indicate number of defects reported at the corresponding site. R.L., Round ligament ; F.T., Fallopian tube ; Ov., Ovary ; U-o.L., Utero-ovarian ligament.

ligament was separated from the rest of the anterior layer of the broad ligament. Bowel which had passed through this aperture remained in the general peritoneal cavity just as in the cases where both layers were involved. In 9 cases the defect communicated with a space between the layers. In 5 of these, this took the form of a peritoneal sac. In 4, including the present case, it is definite that no such sac was present. The edges of the defects have been variously described as ' thin and smooth ' and ' white, scarred, and fibrous '.

6. *The Site of the Defect* (*Fig.* 137).—This is subject to very great variation in those cases where it has been precisely described. It was on the left

side in 9 out of 23 patients, on the right side in 12; bilateral defects were present in 2.

The relationship to the adnexa was as follows: medial to and below the utero-ovarian ligament in 3 (Fagge, 1917; Hunt, 1934; Masson and Atkinson, 1934); between the utero-ovarian ligament and the tube in 6 (Fagge, 1917; Dunn, 1926; Hunt, 1934; Stimson, 1928; Caplan, 1930; and Cooper, 1931); between the tube and round ligament in 2 (Janes, 1929; present case); below the round ligament in 2 (Janes, 1929; Reineke, 1937); through the meso-ligamentum teres in 2 (Pidcock, 1924; Goode and Newbern, 1944); through the suspensory ligament of the ovary in 1 (Shephard and Waugh, 1942).

Table I.—SHOWING THE MAIN FEATURES OF THE DEFECTS IN REPORTED CASES

SITE OF DEFECT	NATURE OF DEFECT	SIDE	AGE	PARITY	AUTHOR
Medial to and below utero-ovarian lig.	In one layer. Peritoneal sac present	Left	61	V	Fagge (1)
Between utero-ovarian lig. and tube	In one layer. Peritoneal sac present	Right	49	0	Fagge (2)
? Between utero-ovarian lig. and tube, emerging between tube and round lig. in front		Right	?	?	Cooper
Beneath round lig.	In one layer. No peritoneal sac	Left	34	V	Pidcock
Between utero-ovarian lig. and tube	In both layers	Bilat.	44	V	Dunn
Below round lig.	In both layers	Left	58	?	Janes (1)
Between tube and round lig.	In one layer. No peritoneal sac	Left	36	VII	Janes (2)
Between utero-ovarian lig. and tube	In both layers. No viscus involved	Right	60	III	Hunt (1)
Beneath tube, round lig. and utero-overian lig.	In one layer. No peritoneal sac	Left	25	II	Hunt (2)
Between ovary and uterus ½ in. below tube	In both layers	Right	56	?	Caplan
Medial to and below utero-ovarian lig.	In one layer. Peritoneal sac present; ovary in aperture	Right	?	I	Masson and Atkinson
Beneath round lig.	In one layer. No peritoneal sac	Left	42	III	Goode and Newbern
Beneath tube and round lig.	In both layers	Right	48	?	Reineke
"Right tube and round lig. formed sharp upper inelastic boundary."	In both layers	Right	30	0	Stimson
"In para-uterine sulcus"	In one layer only. Peritoneal sac present	Bilat.	18	?	Hermann
?	In one layer only. Peritoneal sac present	Left	?	? (Multip.)	Dorman
?	In one layer only. No peritoneal sac	Right	?	?	Finlaison
?	?	Left	56	II	Füssel
Into suspensory lig. of ovary	In both layers	Right	25	III	Shephard and Waugh

Three theories have been advanced and repeated to explain the occurrence of spontaneous defects in the broad ligament. The first regards them as congenital anomalies; the second as the result of previous inflammation in the neighbourhood of, or between the layers of, the broad ligament; and the third as the result of trauma, especially that associated with child-bearing.

It seems clear that no one theory is adequate to explain all the cases. In Quain's case there was clear evidence of old inflammatory adhesions between the broad ligament and the mesentery. Finlaison (1946), alone of subsequent observers, has reported a case in which there was evidence of previous pelvic inflammation, and most writers have commented upon its absence. Developmental error can explain the rare occurrence of the condition in nulliparous women, but there are many difficulties in the way of its application to all cases. The defects show great variability in site and type and the symptoms usually arise in the middle years of life. In the one case where the edge of the defect was submitted to microscopical examination (Honsell, quoted by Füssel, 1937), it was found to consist of "typical scar tissue". Further, Caplan (1930) described a defect in a patient of 56, and stated that

it had not been present when a laparotomy was performed three years previously. All these findings are more readily explicable as the results of trauma. Direct trauma from without is inconceivable without damage to neighbouring and overlying structures. It may be noted in this connexion that Finlaison's patient had undergone an operation for appendicitis eighteen months previously, and a drainage tube inserted then was thought to be the cause of her defect. Thrust from normal intestinal movement or sudden local expansion of the gut, seems by itself an improbable cause from the nature of the organs concerned, and is rendered still less likely by the finding of ovarian migration into the aperture (Masson and Atkinson, 1934) and of defects without involvement of any viscus.

Pregnancy must be given special consideration in view of the fact that 80 per cent of the patients have borne several children. During pregnancy the broad ligament is under tension, and it is not difficult to conceive that some minor trauma at this time may result in a small tear in one or both of its layers; continual tension holds the wound edges apart and prevents normal healing. By the time it ceases to act these edges are covered with serosa. However, this concept cannot explain the occurrence of true sac formation reported in 5 patients, 2 of them nulliparæ. The facts here can only be reconciled by assuming that there may sometimes be a pre-existing weakness in the ligaments, which under relatively minor degrees of stress gives rise to a saccular defect. A similar condition is thought to account for the saccular type of congenital diaphragmatic hernia.

Diagnosis.—This is unlikely as a rule to be made accurately before operation. When the strangulation is not severe, and when the condition is right-sided, it may mimic acute appendicitis closely. The possibility of intestinal strangulation in a broad ligament defect is worth bearing in mind when a middle-aged multiparous woman is seen suffering from acute abdominal disease, as the condition calls for operative intervention.

Treatment.—Urgent laparotomy has usually been necessary, though a few cases have been relatively subacute and operation has been delayed for a day or two. Some operators have left the defects unrepaired. It seems safer to close them to prevent recurrence, though there is no record of a patient having suffered twice.

I wish to thank Prof. Ian Aird, Professor of Surgery at the Post-Graduate Medical School, for his encouragement and helpful criticism in the preparation of this paper.

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RUPTURED QUADRICEPS

A CLINICAL STUDY

BY ERIC I. LLOYD, LONDON.

RUPTURE of the quadriceps femoris has several peculiarities. It almost always occurs in heavy, elderly men from indirect violence, it is often wrongly diagnosed, and there is probably rather a high risk of pulmonary embolism.

CAUSE

The injury is nearly always associated with stairs, and the most usual story is of a descent in the dark with a missed foothold followed by a sudden convulsive extension of the knee which is not bearing weight. A fractured patella might result in a younger man, but presumably the degenerative changes of advancing years weaken the tendon and this generally breaks near its insertion. Fracture of the patella is probably about eighty times as common as ruptured quadriceps, but the latter is in its turn a good deal more frequent than rupture of the ligamentum patellæ, of which the present writer has seen only one example.

DIAGNOSIS

This would be easy if the condition were always remembered as a possibility. Inability to extend the knee associated with a palpable depression in the surface of the extensor tendon can only be due to

one thing but the diagnosis is often missed owing to lack of familiarity (*Fig.* 138). The following are three examples :—

1. A man of 77 had " fallen down some steps and doubled up his left leg " five or six years previously. He saw a bone setter who " treated him for dislocation of the patella ", but there was an old undiagnosed rupture of the quadriceps and the patient had been lame ever since the accident.

2. Another man of 77, who weighed 16 st., caught his heel and slipped when coming downstairs. Both quadriceps were ruptured, but operation was delayed because a neurologist diagnosed hæmatomyelia of the spinal cord and the young surgeon had never previously seen a case of ruptured quadriceps.

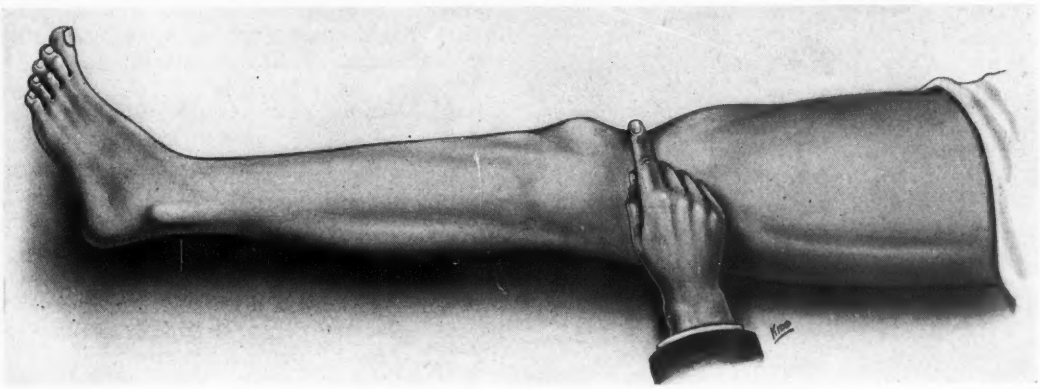

FIG. 138.—A depression can be felt in the extensor tendon.

3. An alcoholic gentleman of 63 had a comedy accident three weeks before being seen. He fell from a step ladder and then took two paces backwards on to a bucket which gave way under him so that he twisted his knee very badly. He remembered having to force it straight and had much bruising and a joint full of fluid when he was examined. Treatment was prescribed for an acute synovitis with effusion, but he subsequently slipped again and then it became clear that he had a ruptured quadriceps which may have been present all the time. He later consulted another surgeon, who sutured his tendon several months after the injury.

Confirmation of the diagnosis is rarely necessary when once it is suspected, but further evidence can be obtained from an anteroposterior radiograph of both knees on one film. The patella on the ruptured side is quite obviously lower than the normal and this is caused by loss of postural tone, which ordinarily exerts traction on the patella and ligamentum patellæ.

A divided muscle retracts and the only possible natural repair is by organization of the resulting hæmatoma with a long fibrous union across a gap of about 2 in. This greatly weakens the muscle and causes permanent lameness so that operative treatment offers the only chance of regaining normal power. The operation is not, however, of extreme urgency and there is ample time for a routine skin preparation and estimation of the blood-urea and blood-pressure.

Twenty-four hours before the operation a 3½ in. ' viscopaste ' (zinc-gelatin) bandage is fixed to the foot from the web of the toes to a point 2 in. above the ankle-joint, and the upper inch of this is then covered by a cuff of felt 4 in. wide which is stitched or stuck round the limb (*Fig.* 139). 500,000 units of penicillin are also given intramuscularly in one dose on the day before operation and this is repeated next day.

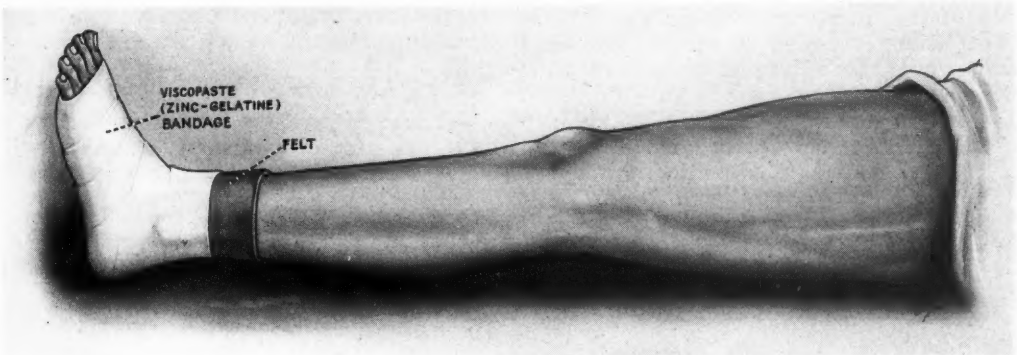

FIG. 139.—Pre-operative treatment.

TREATMENT OF RECENT CASES

This should be operative in practically all recent cases and a general anæsthetic is necessary. Hæmorrhage is very slight and it is much better not to use a tourniquet because of the risks of vascular stasis in these old patients. An Esmarch's bandage is even worse, because quite severe shock can be caused by driving the products of tissue damage into the circulation. A generous curved incision, concave

proximally, exposes the frayed torn ends of the tendon and surrounding blood-clot which generally lies inside the knee-joint and which should be removed. The synovial membrane may be sutured at this stage, but it is by no means an essential step

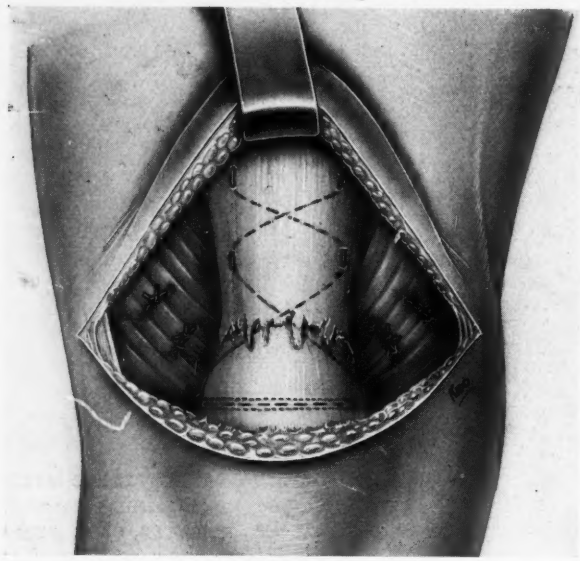

FIG. 140.—Stay suture passing through transverse hole in patella supplemented by interrupted sutures on either side.

in the operation and may be omitted if bruising and œdema of the tissues make it difficult.

Sufficient tendon occasionally remains attached to the patella for direct suture to be possible, but it is much more common to have to drill the bone and to proceed as follows: A suture of No. 1 kangaroo tendon is inserted through the tendon as shown in *Fig.* 140 and is then passed through a single transverse drill hole in the upper third of the patella. This stay suture re-establishes continuity of the rectus femoris with the patella, but it must be supplemented by a series of interrupted chromic catgut sutures both centrally and on either side before the wound is closed without drainage.

A strong plaster slab is now placed along the back of the limb so that it rests on the felt strip just above the ankle and upon a second which encircles the upper one-third of the thigh. (*Fig.* 141.) The plaster slab is held to the limb by a few turns of plaster bandage above and below the field of operation and an ample dressing is firmly bandaged over the wound so that it can later be removed without disturbing the plaster which holds the knee-joint in full extension.

TREATMENT OF OLD OR NEGLECTED CASES

Suture is generally worth while up to three months but the operative details differ because it will usually be found impossible to approximate the divided ends of the tendon. They should then be drawn together as much as possible in the way previously described and the resulting gap bridged by fascia lata or peroneal tendon derived from half the thickness of peroneus longus.

POST-OPERATIVE CARE

The patient is returned to bed with the injured limb on a pillow beneath a cradle. As soon as he has regained consciousness he sits up and is provided with a back-rest and overhead hand-hold, which he is encouraged to use frequently for changing his position.

He is asked to move his ankle and toes through a full range of movement for a few minutes every hour and he can quite easily do this in the plaster described, which was recommended by Böhler for cases of fractured patella and which allows complete freedom of the ankles, toes, and hips.

PULMONARY EMBOLISM

These patients are frequently over 70 and there is a real risk of pulmonary embolism if they are allowed immobility. The late Sir Bernard Spilsbury believed that the commonest starting-point is stagnation in the venous plexus of the calf and that the most efficient way of emptying it is by active muscular contraction.

Example: A man, aged 83, and weighing 14½ st., was operated upon five days after he had ruptured his quadriceps. He did well until the fifth day, when he

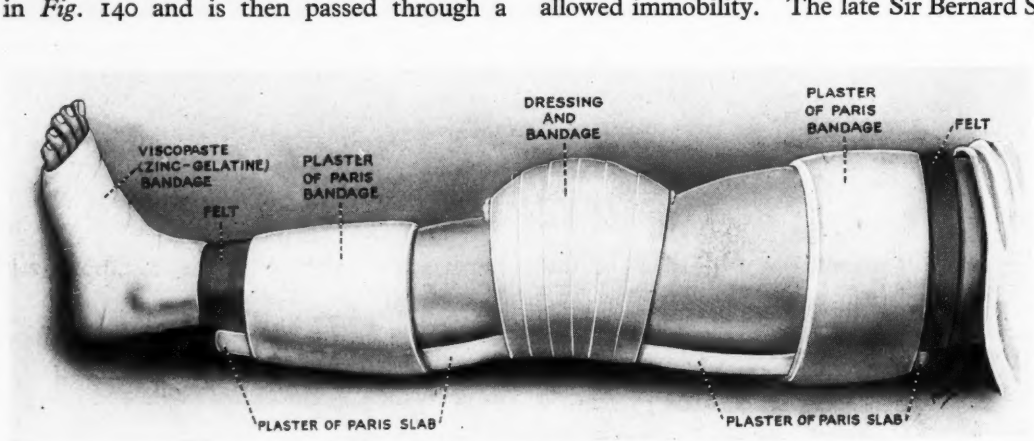

FIG. 141.—Method of treatment at the close of operation.

complained of feeling giddy: He died in a few minutes from pulmonary embolism, which was thought at the post-mortem to have started in the deep femoral vein. No special precautions against thrombosis had been taken, and it is the only occasion in this series where the records state that a tourniquet was used during the operation.

AFTER-TREATMENT

In four or five days the patient may begin static exercises to his quadriceps and he is gradually coaxed into moving his plastered leg and even lifting it off the bed. This is a valuable way of diminishing the risk of adhesions at the site of rupture and is probably a great help in the regaining of flexion when the plaster is later discarded.

The stitches are removed in ten days and the plaster completed in such a way that static contractions of the patella remain possible. This can be done by putting a vaselined cotton-wool pad just proximal to the patella and removing it through a lateral hole as the plaster dries.

The patient should be out of bed and walking with a stick next day and soon afterwards be taking quite long walks out of doors with full weight-bearing. The plaster is removed six weeks after the operation and active and passive movements with radiant heat are given to the knee-joint.

RESULTS

One patient aged 77 had acquired 70° of active flexion by his own unaided efforts on the ninth day after removal of the plaster, but it is more common in the elderly to find some difficulty in regaining full movements rapidly. The late cases in which suture has been performed several months after the accident have usually some limitation of movement of the knee-joint and are less satisfactory than the recent cases. Bridging a gap with tendon or fascia inevitably diminishes the efficiency of the quadriceps mechanism and is likely to cause loss of the last 5–10° of active extension and a resulting slight insecurity in walking. There may also be some loss of flexion, but this is no great disability if the knee bends to a right angle.

SUMMARY

A clinical survey of examples of ruptured quadriceps indicates that the diagnosis may be missed from unfamiliarity with the condition.

Suggestions are made for the management of cases and for the avoidance of certain complications.

The results of operative treatment are concluded to be excellent in the recent examples and fairly satisfactory in those operated upon within three months of the injury.

RUPTURE AND SUTURE OF FEMORAL VEIN

By CHARLES WELLS, Liverpool.

The generally practised operation for varicose veins involves ligation and division of all the recognizable radicles and retrograde injection of the saphenous tract with a sclerosing agent. The internal saphena is then divided and tied near its junction with the femoral vein.

Some time ago I came into an operating theatre when a junior had just completed such an operation. Unfortunately, having divided the branches and injected the main stem, he had had an accident, as a result of which the femoral vein had been torn at the point of entrance of the internal saphenous. Torrential venous hæmorrhage had followed and with some difficulty he had succeeded in tying the femoral vein above and below the tear. He had then sutured his wound.

The limb was an unhappy sight—swollen and dusky, with the injected veins distended and black. After a brief consultation we decided to reopen the wound and see what could be done.

The femoral vein, distal to the ligature, was distended to the diameter of an index finger with extremely dark blood under great pressure and there was clearly no adequate venous return from the limb (*Fig.* 142). The tension was such that rupture appeared to be only a matter of time. If this had occurred it would have been calamitous. So far as we could judge, ultimate loss of the limb was not improbable.

Arterial clamps were available and one was applied to the common femoral artery without any noticeable change in the tension in the vein. Other clamps were then applied to the vein above and below the ligatures and the latter cut away (*Fig.* 143). Some blood and clot escaped. We were then faced with the problem of repair.

As no special suture material was available, we used fine chromic catgut mounted in an eyeless needle and repaired the vein by longitudinal suture, after irrigating the exposed parts with citrate solution.

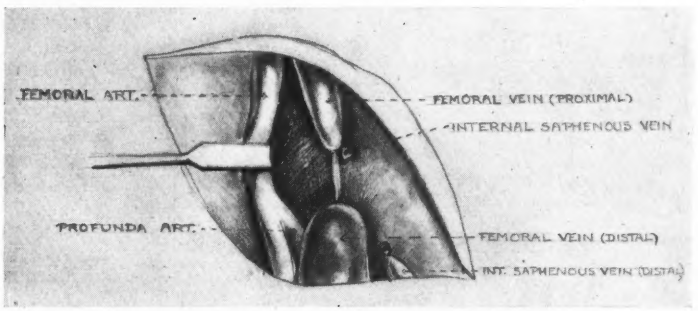

Fig. 142.—Showing the condition on reopening the wound.

We then removed the clamps, only to find we were back where we started. The suture-line had completely obliterated the lumen of the vein.

After re-applying the clamps, the suture was taken away and the ligature on the stump of the saphena was removed, for the first time. This afforded a scrap more tissue, but there was still very little to bridge the gap and we debated the

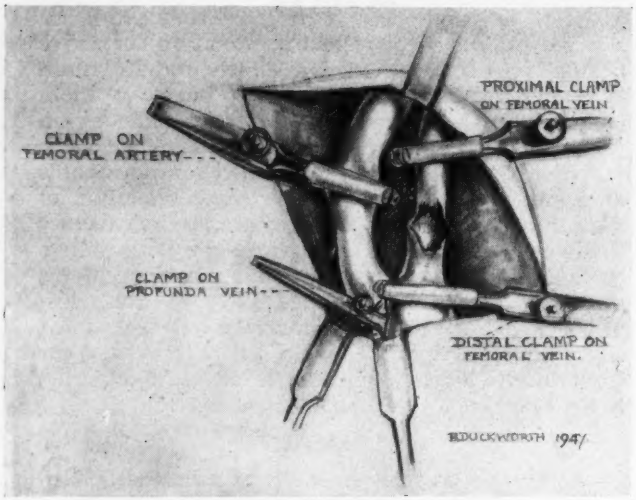

FIG. 143.—The clamps applied and the ligatures removed showing the tear in the femoral vein.

various possibilities—anastomosis over a vitallium cuff, a free vein graft, a free graft supported on a cuff, etc. Finally we decided upon transverse suture with similar chromic gut and this was done with a simple running-suture, beginning from the centre for the sake of symmetry and working outwards in two lengths to make a single-layer repair. With the removal of the clamps we were amply rewarded for a long and tedious job.

As the congestion subsided there was minimal leakage from the suture line and this was easily checked with gentle pressure for a few minutes.

Immediately upon returning to the ward, an intravenous saline drip containing 5000 units heparin per 540 c.c. was set up in one arm and was maintained for three days, after which the amount of heparin was halved and the same drip run for a further three days. At this time a blood-stained collection of an ounce of serum evacuated itself from the wound. The patient left hospital on his own feet eleven days from operation and has remained well.

SHORT NOTES OF RARE OR OBSCURE CASES

FOREIGN BODY IN THE ABDOMEN

By N. J. NICHOLSON

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CASE REPORT

G. B., aged 61, was referred on account of osteo-arthritic changes in both knees. In the course of a routine history the patient said that he had undergone an operation in another hospital on May 17, 1927, by a surgeon who is no longer alive. On Oct. 25, 1930, he passed by rectum the glass tube seen in the photograph (*Fig.* 144). The

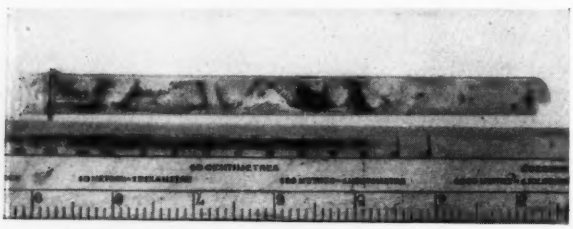

FIG. 144.—The drainage tube as passed by the patient.

operation was performed for perforated duodenal ulcer. The patient produced a written copy of his operation notes which he had made surreptitiously whilst in hospital. This showed that the ulcer was infolded in two layers and covered with omentum after which a posterior no-loop gastro-enterostomy was performed. He recalled that a tube was present in the lower abdomen, of glass, to which a rubber tube was attached. In subsequent dressings the tube was twisted or taken out and replaced. Presumably this applied to the rubber part only. The process was very painful, but eventually "it healed up wonderful". Later whilst still in bed, he suffered much from constipation and had enemata every other day, as well as liquid paraffin and a good deal of scolding. Seven weeks after operation he was allowed up and passed stools then no thicker than an earthworm—in spite of Epsom salts as hot as he could drink. A doctor made a rectal examination and attributed the constipation to adhesions. He suffered also from pains in the legs which "clicked and twitched".

After leaving hospital he had pain in the right side of the perineum, which made him unable to ride in a car or a bus. He would walk with his hands in his trouser pockets to press the lower abdomen to obtain some relief. After six months he was passed fit for work in spite of his objections. Later the pain became less, but returned occasionally in a violent colicky form which made him roll on the floor and rub his body. He vomited sometimes and sometimes passed blood from the bowel, but not a great deal. One day he passed the glass tube whilst at stool.

At the request of the writer the patient brought the tube at his next visit. The tube is 6½ in. long. One end is slightly broken and sharp. It has a number of small holes in it like a Carrel-Dakin tube.

The patient had never washed the tube, which still contains a lot of dry fæces adherent for the most part to the sides of the tube but with powdery fæces and small dry lumps still present in its lumen.

Since the passage of the tube he has had no further ill effects.

Comment.—It is interesting to speculate when the tube ulcerated into bowel; possibly it had done so at the end of seven weeks, when the very narrow stools were seen. The pain in the legs may have been due to pressure from the foreign body on the lumbosacral plexuses.

COMPLETE CONGENITAL ABSENCE OF THE VAGINA ASSOCIATED WITH BILATERAL HERNIÆ OF UTERUS, TUBES, AND OVARIES

By G. RUSSELL THOMSON
EX S/LDR., R.A.F. MEDICAL SERVICE

ALTHOUGH abnormalities of the genital tract are comparatively common, and frequently described, complete congenital absence of the vagina, associated with bilateral herniæ of uterus, tubes and ovaries, appears to be more in the nature of a rarity, as only one similar case has been discovered in the literature. It seems worth while putting this case on record.

CASE REPORT

HISTORY.—On August 15, 1946, a W.A.A.F., aged 20 years, was admitted to P.M., R.A.F. Hospital, Halton, complaining that she had never menstruated. She mentioned that she had occasional twinges of pain in the left inguinal region for the past three years, this being attributed to a small hernia for the repair of which she was already on the waiting list of a hospital.

Past History.—The non-appearance of her periods at the age of 15 years began to worry her, and after consultation with her doctor, her tonsils were removed! This being ineffective, she was then treated with stilbœstrol and thyroid extract, but still without effect. Some months after entry into the W.A.A.F. she was examined and found to have an absent vagina.

Family History.—The patient was unaware of any congenital abnormality in her own family.

ON EXAMINATION.—On admission, the patient was outwardly normal, being typically effeminate, bright, and cheerful. The breasts were well developed, and the pubic hair normally distributed. She was rather more plump and stocky than the average, but nevertheless quite feminine in build. On Aug. 17 she was examined under an anæsthetic. The labia majora and minora, clitoris, and urethra were normal, but there was complete absence of the vagina, and apparent absence of uterus and ovaries.

FIRST OPERATION.—On Oct. 12, a new vagina was constructed by McIndoe's method. Her convalescence was uneventful, and she was discharged well from hospital, with instructions to return for observation, and to have her hernia repaired.

SECOND OPERATION.—On May 5, 1947, she was readmitted for repair of the left inguinal hernia. Preoperative examination revealed a globoid movable mass in the left inguinal region, anterior to the superficial ring, and closely simulating a hernia. A similar mass was then found in the right inguinal region. In view of the tenderness and mobility of the mass, a further congenital abnormality was suspected, with the possibility of the ovaries being in an abnormal, but not impossible, position.

At the operation, on May 20, a left inguinal incision down to the external oblique muscle displayed a mass projecting through the external ring and too large to be returned within the inguinal canal. The external oblique was divided and an obvious hernial sac opened. It revealed within a normal ovary, Fallopian tube with fimbriated extremity, and a rudimentary uterus plastered on the extraperitoneal aspect of the posterior wall of the sac, and fading away into the neck of the sac, as a tubular band (*Fig.* 145). The Fallopian tube entered the uterus, and the fimbriated extremity lay free and curled round the ovary. The contents of the sac were returned to the abdominal cavity and the neck of the sac closed. The conjoined tendon was sutured to the deep aspect of the inguinal ligament and routine closure of the wound completed.

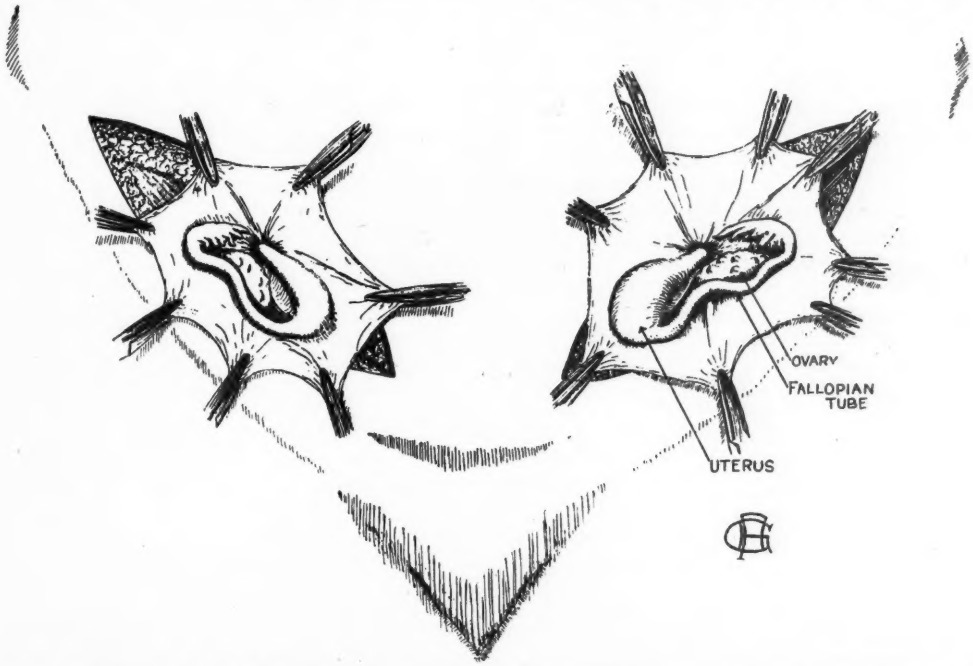

FIG. 145.—Diagram showing right and left inguinal hernial sacs opened, with ovary, Fallopian tube, and uterine muscle present in each.

A similar abnormality was found to exist on the right side—the uterus being represented here, however, as a much smaller plaque of tissue plastered on the sac wall, with the end of the Fallopian tube buried in it (*Fig.* 145). The fimbriated extremity and the ovary were normal. The abnormality was dealt with in a similar way to the left side.

PATHOLOGY.—Sections taken at operation confirmed the presence of uterine muscle on both sides.

Convalescence was uninterrupted and she was well when discharged from hospital.

DISCUSSION

Reference to literature shows few cases comparable to the one described. D. Deutschman (1923) describes the almost identical case of a Russian woman of 34 years, of typically feminine appearance, complaining of bilateral inguinal pain intensified on exertion. At operation, ovaries and Fallopian tubes were found in the inguinal herniæ, and were easily reducible into the inguinal canals. There appeared to be no uterine tissue in the left sac, but a rudimentary uterus was found in the right. In addition, there was absence of the vagina.

H. H. Young (1937) describes a case of true hermaphroditism in which the external genitalia consisted of a normal penis and right testicle, the left testicle missing. In a left inguinal hernia sac was found the uterus, left ovary, and Fallopian tube.

Francis Stock (1944) describes the case of a Yomba girl of 20 years, who had never menstruated and showed complete absence of the vagina, being normally feminine otherwise. During the operation to form a new vagina by Baldwin's method, "it was seen that across the pelvic floor immediately behind the bladder was a narrow tube 3 in. long with bulbous extremities. From the extremities there ran normal round ligaments and short Fallopian tubes. The ovaries were present and normal and the fimbriated ends of the Fallopian tubes curved round them in the normal manner".

Normally the uterus is formed by the fusion of the two Müllerian ducts. The latter are solid at first, canalization taking place later from below upwards.

Various causes of failure of union of the two Müllerian elements have been described, none of which accounts for all cases. Von Winckel gives two possibilities which might occur: (1) A vesico-rectal peritoneal fold of vascular origin may stretch across the pelvis from front to back and thus prevent union. (2) The Müllerian ducts may be held apart by abnormally short round ligaments.

In the case described it is assumed that there has been failure in the development of the Müllerian ducts, their lack of fusion resulting in absence of the vagina. Development of the uterus has also been interfered with, the uterus being represented by plaques of uncanalized uterine muscle on either side. The descent into the inguinal canals is interesting. Deutschman (1923) points out that normally the ovary descends from the lumbar region in the eighth week to its final position in the true pelvis. In his case he considers the ovary descended to the inguinal canal, dragging along with it the rudimentary uterus and tube on the right side, and the tube on the left side.

Presumably a similar occurrence has taken place in the case described here, with the added abnormality of finding uterine muscle present in the right as in the left inguinal canals.

I wish to thank the Director-General of the R.A.F. Medical Service for permission to publish this report; and also Dr. Geoffrey Fisher for the diagram.

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MALIGNANT DEGENERATION OF AN ANAL FISTULA

BY F. S. A. DORAN

SURGICAL CHIEF ASSISTANT, MANCHESTER ROYAL INFIRMARY

ALTHOUGH this condition is well recognized, and examples are included in standard works on the surgery of the rectum and anus (Gabriel, 1937), it is still regarded as something of a curiosity. Yeomans (1929) reported 3 cases, in each of which malignancy developed in the scar resulting from the operation. Reviewing the literature upon this subject, Rosser (1931) was only able to add a further 4 cases; but he was able to report another 5 from his own practice and to add 2 more rather dubious ones. To these must be added the 2 instances of carcinomatous degeneration taking place in anal fistulæ published by Ducassi and Smith (1945). It is pointed out by these writers that a fistula should not be considered as having undergone a malignant degeneration unless it is certain that no tumour exists in the anal canal or rectum, and that the internal opening of the fistula does not lie in malignant tissue. Histologically, 5 of the reported cases were adenocarcinoma, the remainder being composed of colloid carcinoma, epithelioma, and 1 basal-cell carcinoma.

CASE REPORT

A female patient, aged 65, was admitted to the hospital in May, 1947, complaining of soreness and a continual discharge from the region of the anus. She gave the history that a year previously she had developed a 'boil' near the anus which had burst and discharged small amounts almost continually ever since.

On examination it was found that on the right-hand side of the anus, and in no way involving it, there was a firm, sessile, warty, papillomatous mass 1 in. in diameter. There was no evidence of any tumour in the rectum and, as has been implied, the anal canal

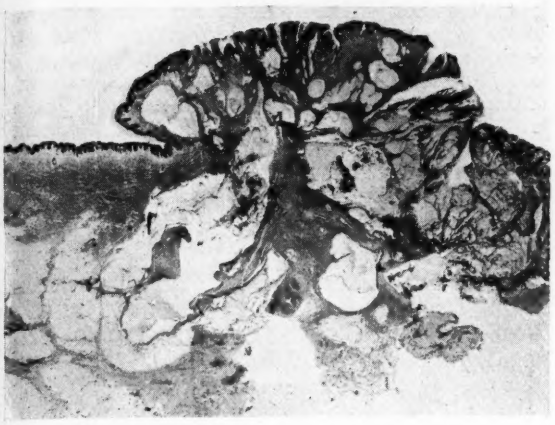

FIG. 146.—Low-power section of the specimen.

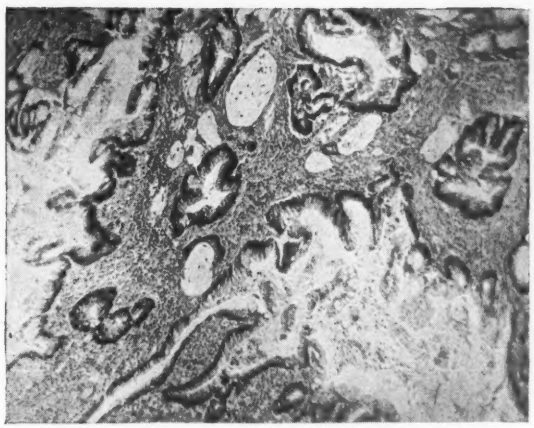

FIG. 147.—Higher-power section of the specimen, showing the cellular arrangement of the adenocarcinoma and colloid degeneration.

was free from any neoplastic change. The internal orifice of the fistula could not be identified.

The mass and a large wedge of skin and underlying connecting tissue were excised with the diathermy and sent for section. The histological report showed that the mass was an adenocarcinoma, growing in parts in colloid form, ulcerating through the skin, and extending into the subcutaneous tissues (*Figs.* 146, 147).

All the reported cases have occurred in patients of the cancer age, the youngest being 48 years. It is evident, however, that although most of the cases have occurred in chronic fistulæ of long standing, it can occur after the fistula has only existed for a year, as happened in one instance reported by Rosser (1931) and as the case that has just been described shows.

I wish to thank Mr. R. L. Newell, Honorary Surgeon to the Manchester Royal Infirmary, for his help in the preparation and presentation of this case report.

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A CASE OF NEURINOMA OF THE VAGUS NERVE IN THE NECK

BY R. S. MURLEY

TUMOURS arising in the cervical portion of the vagus nerve are of sufficient rarity to warrant publication of this case.

The type of tumour here described arises from the neurilemma sheath and is innocent in character. Although this type of tumour is usually called a neurinoma in this country, in the United States it is more commonly described as a 'neurilemoma'. Further titles less frequently used are 'Schwannoma', 'perineurial fibroblastoma', 'peripheral glioma', and 'neurofibroma'. The latter term is, however, a misnomer for this type of tumour.

CASE REPORT

HISTORY.—Miss Z. A., aged 19, a children's nurse, was admitted to St. Bartholomew's Hospital on May 24, 1947. Three years previously she had first noticed a lump in the right side of her neck, which had gradually increased in size. During the five months before admission she experienced occasional aching pain in the swelling which radiated to the shoulder and mastoid region. A few weeks before admission she was referred to the Brompton Hospital on account of a mild cough and some loss of weight. Investigation there revealed no chest disease, but a diagnosis of tuberculous lymphadenitis was made and she was then referred to St. Bartholomew's Hospital.

ON EXAMINATION.—The patient appeared perfectly healthy apart from her local condition and she was apyrexical. Examination of the neck revealed a smooth, firm, ovoid swelling, measuring some 4 cm. in its vertical and 3 cm. in its horizontal axis. It was situated below and behind the angle of the jaw, anterior to the sternomastoid, and in the line of the carotid sheath. It could be moved across the line of the carotid sheath but not vertically, and could not be transilluminated. No glands were palpable in the neck and the throat was healthy.

Plain radiography of the chest was normal: the erythrocyte sedimentation rate was 11 mm. in the first hour: total white-cell count 7800, and differential white-cell count normal.

A clinical diagnosis of tuberculous lymphadenitis was made.

AT OPERATION.—This was performed on May 27 under nasal endotracheal gas–oxygen–ether anæsthesia. A skin-crease incision was made overlying the centre of the tumour and, as soon as the deep fascia was incised, a slightly enlarged tonsillar lymph-gland was seen. Beneath this there was a further swelling situated within the carotid sheath, with the internal jugular vein spread out over its surface and displaced backwards and laterally. On opening the carotid sheath it was seen that the tumour was incorporated in the vagus nerve, with the internal carotid artery lying medial and anterior to it. The tumour extended to just below the base of the skull and was roughly in the position of the nodose ganglion of the

vagus: close to its upper extremity there was a branch of communication to the superior ganglion of the sympathetic chain, while the hypoglossal nerve emerged from behind the tumour and passed across its lateral surface.

Although a thin strand of nerve-fibres could be seen coursing over the lateral aspect of the tumour, it was not thought advisable to attempt enucleation and leave the

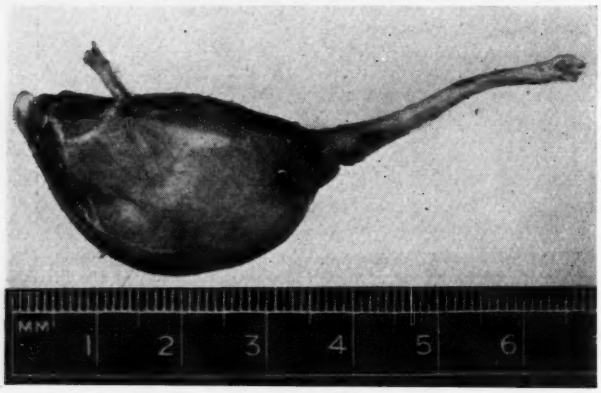

FIG. 148.—Photograph of the neurinoma.

nerve-fibre intact, as the exact nature of the tumour was in doubt. Accordingly, the vagus was divided just above the tumour and again a short way below it; the branch to the sympathetic chain was also divided, and the tumour was removed. The incision was closed without drainage.

THE SPECIMEN (*Fig.* 148).—This consists of an ovoid tumour measuring 3·5 × 2·3 × 2·0 cm., with a short stump of nerve at one end and some 3 cm. of nerve at the other end. Nerve-fibres can be seen thinned out

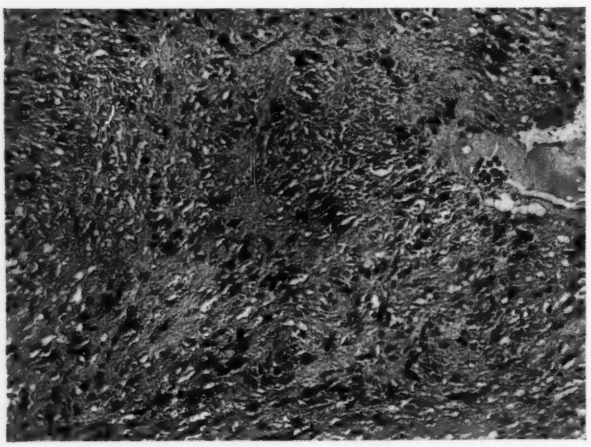

FIG. 149.—Microphotograph of a section of the tumour (*see* text).

over the surface of the specimen with the short branch of communication to the sympathetic chain attached near its upper end. On section the tumour is seen to be completely encapsulated, firm in consistency, and yellowish in colour, with areas of red mottling.

Histologically (*Fig.* 149), there are irregularly arranged bundles of collagen interspersed with œdematous patches and occasional hæmosiderin deposits. The tumour nuclei in some parts are palisaded, and there is a well-developed fibrous capsule with compressed nerve-fibres derived from the attached nerve. The appearances are those of a neurinoma.

SUBSEQUENT PROGRESS.—Immediately following operation the patient had a husky voice due to paralysis of the right vocal cord. There were no other features attributable to vagus nerve section and progress was uneventful, the patient being discharged on the tenth day after operation. Speech therapy was instituted in hospital and was continued for two months after the patient's discharge. She was examined at intervals and when last seen, six months after operation, there was still some huskiness of the voice, which causes her no trouble and which is not entirely without its attractions.

DISCUSSION

Other cases of neurinoma of the vagus occurring in the neck have been published by Cutler and Gross (1936), Sekiguchi and Oije (1926), Serafini (1931), and Mayo and Barber (1934). These articles refer to only 4 definite cases and a further 3 doubtful ones. Cutler and Gross (1936), publishing 25 cases of benign and malignant growths of the peripheral nerves unassociated with von Recklinghausen's disease, describe 2 definite examples of neurinoma of the vagus nerve in the neck. In 2 more of their cases they could not definitely identify the site of origin of the tumour, although it seemed to be closely associated with the vagus. They, like other authors, stress that these tumours are not uncommon in the neck, although in the majority of cases they arise from the cervical nerve-roots and sympathetic chain.

The most complete publications on this subject are those of Stout (1935, a, b). Stout published some 50 fresh cases of 'neurilemoma' with no instance of vagus nerve involvement; he found only 194 additional cases in the literature at that time and doubted whether the tumour was so uncommon as this would suggest, although he confirmed the rarity with which cranial nerves in the neck are the site of such tumours. The hypoglossal nerve appears to be even more rarely involved than the vagus, and the writer has only found 2 cases in the literature, one published by Mayo and Barber (1934) and the other by Friedman and Eisenberg (1935).

Preservation of nerve function is a consistent feature of these tumours, whereas disturbance of nerve function, more rapid growth, and infiltration of the surrounding structures is characteristic of the much rarer malignant types of tumour.

Stout (1935, b), reviewing the literature, found no record of a malignant tumour of the vagus, although he refers to a personal case of malignant paraganglioma of the nodose ganglion, which he does not regard as a true nerve tumour. Since then, Furber and Fox (1940) have published a case of perineural fibrosarcoma of the vagus.

With regard to the management of tumours of the cranial nerves in the neck, there can be no doubt that in the innocent cases the treatment of choice is enucleation of the tumour with preservation of the nerve. Where the tumour is obviously or suspiciously malignant, then the affected part of the nerve should be removed too.

At the time of operation in the case here described the writer did not appreciate that he was dealing with an innocent tumour which could be enucleated, and the nerve was therefore sacrificed. This was a mistake, and an attempt should first have been made to shell out the tumour after incising its capsule. From examination of the specimen it seems probable

that enucleation could have been effected quite readily.

A further lesson to be derived from this case is that it is an unwarranted assumption to conclude that a laterally situated tumour in the neck is a diseased lymph-gland merely because this is a common situation for such a swelling. In this case, the history of slow and steady enlargement over a period of three years, the absence of any apparent primary infective focus, and the lack of tenderness should, perhaps, have encouraged consideration of alternative diagnoses.

SUMMARY

A case is described of neurinoma of the vagus nerve in the neck, occurring in a woman of 19. The clinical diagnosis was that of tuberculous lymphadenitis.

The tumour was excised together with a segment of the parent nerve, but subsequent examination suggested that enucleation of the tumour with preservation of the vagus nerve would have been the treatment of choice.

Apart from huskiness of the voice, there have been no ill effects from removal of a portion of the right vagus nerve in the neck.

Reference to the literature shows that although neurinomata are not unusual in the neck, they most commonly arise from the cervical plexus. Only 4 definite cases, and a further 3 doubtful cases, of vagus nerve neurinoma have been found recorded.

I am indebted to Dr. George Cunningham of the Pathology department, St. Bartholomew's Hospital, for his report on the tumour and for the photomicrograph; to Mr. Norman Harrison for the photograph of the tumour, and to Mr. John Hosford for permission to publish this case.

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LARGE ULCERATING SEBACEOUS CYST OF FOREHEAD

BY A. C. LOVETT-CAMPBELL, LONDON

CYSTS of this character are not notably prevalent in Africans, whose amply exposed skins, unrestricted by excessive clothing, generally maintain unimpeded secretion.

In certain tribes, however, who have adopted heavy variegated head-dress, and particularly among their women folk, who swathe their foreheads with yards of none too clean linen and cotton fabrics, retention cysts are apt to develop.

This Hausa girl from Northern Nigeria had a smooth cyst on the forehead, which seven years later attained to the size and condition shown in the first photograph (*Fig.* 150).

Surface ulceration had broken the skin extensively. The tumour hung from the forehead by a pedicle that had undergone considerable elongation through continuous traction.

She suffered from severe neuralgia of the scalp.

Removal was easy by division of the pedicle, after infiltration with 2 per cent novocain (*Fig.* 151).

FIG. 150.—Photograph showing the size and position of the sebaceous cyst.

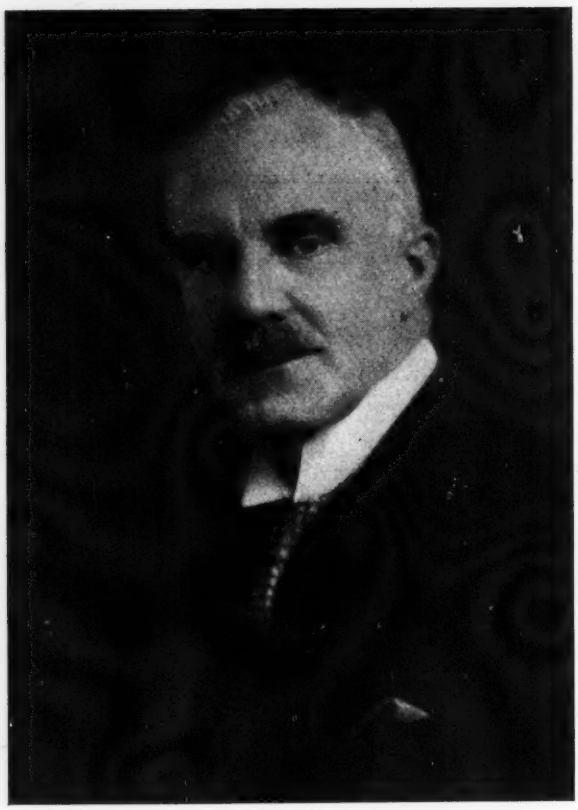
FIG. 151.—The patient after removal of the cyst.

In Memoriam

ARTHUR HENRY BURGESS
(1874—1948)

ARTHUR HENRY BURGESS died suddenly and unexpectedly at the age of 74 while in Edinburgh, where he had gone for the purpose of attending the meeting of the Association of Surgeons in May. His passing without fuss or ostentation was as he would have wished but came as a poignant shock to his many friends.

Burgess was born in 1874 in Manchester and the whole of his life and activities, with the exception



ARTHUR HENRY BURGESS
(1874—1948)

of a period overseas in the war of 1914–18, were associated with that city which he loved so well. For many year he was part of Manchester and no one thought of that centre surgically without the association of his name.

At the outset of his career he spent a year in a shipping office and this training probably inculcated those careful business habits which were characteristic of him. From commerce he went on to Owens College, where he studied Zoology and Physiology. His academic career was brilliant and he won many coveted prizes and other undergraduate distinctions. This period was marked by his obtaining the M.Sc. in 1895, for at that time he thought of embarking on a career as a Zoologist, but circumstances pointed to a promising opportunity in Medicine and in 1896 he graduated with first-class honours as M.B., Ch.B., and in 1899 took the Fellowship of the Royal College. He held a series of house posts, culminating in a couple of years as R.S.O. to the Manchester Royal Infirmary, which was his parent hospital and from then on became the focus of his notable surgical activities for thirty years, though he held other posts.

Whatever work Burgess undertook was carried out to the full limit of his powers, for he always concentrated on the affairs in hand and that was so throughout his very active life.

As surgeon in a teaching hospital he was well placed; he soon made his attributes felt in the Manchester Royal and set both a standard and a pace which many of his associates found rather hard to follow. One of his early accomplishments was the introduction of a stricter and more complete aseptic technique, and the toilet of the theatre under his régime became something of a byword.

As an operator he was quick but thorough, and it was always an education and a pleasure to watch him at work for he was regarded as one of the best technicians of his time. For many years he enjoyed a very large and important consulting practice which was enhanced by his punctuality and personal attention to detail. On the academic side he did notable work for the School in all the usual offices, in due course becoming lecturer and, in 1921, Professor of Surgery and later Emeritus in the School.

In his teaching he concentrated on grounding in principles and essentials and taught his students the importance of doing all ordinary things well.

During the 1914–18 war he went out to Mesopotamia in charge of the Surgical Division of a general hospital and after a time was appointed Consulting Surgeon and was always proud of the fact that his name was inscribed on the mace which was presented to the American College of Surgeons by the Consulting Surgeons of the British Armies. But, his service overseas was not without its price, for he never quite threw off the effects of a severe malaria contracted in Basrah.

His Presidency of the British Medical Association in 1929 was notable for the punctilious way in which he carried out the whole of the duties and obligations of that office. But it was no passing phase, for his interest and realization of the importance of the B.M.A. were sustained throughout his life. In 1925 he was top of the poll in the election for the Council of the College of Surgeons and was proud of his long service on that body. There he had a remarkable record of attendance to all the duties, and during his period as Vice-President it was no mean accomplishment, considering the amount of travel necessitated

from his home in Manchester, that he rarely missed a committee or engagement of any sort.

It was the same on the Editorial Committee of the BRITISH JOURNAL OF SURGERY, which he served for eighteen years. In all these activities his shrewd judgement of men and affairs was valuable, but to the onlooker the constant lesson was the punctilious way in which he carried out the whole of his obligations. Wherever he went he oozed good fellowship and companionship and there was no more popular member of the Moynihan Club, of which at the time of his death he was one of the two surviving original members. He was intensely interested in the technical side of its activities and only a few weeks ago at our meeting in Leeds it was a great lesson to watch him standing for a couple of hours with attention riveted on the details of an œsophagectomy.

In 1933 he was President of the Association of Surgeons.

Burgess was not a voluminous writer, but all that he wrote was founded on large experience and was thoroughly practical. His Murphy Oration delivered in Chicago in 1931, on genito-urinary surgery, the Bradshaw Lecture in 1933 on electro-surgery, and his Hunterian Oration in 1941 were each outstanding. In the first he dealt with Murphy's contributions to genito-urinary surgery. This appealed to him very much for it was an early interest and he had done much to introduce the cystoscopic era into the Manchester School. His Bradshaw lecture, summarizing the knowledge of electrosurgery, showed that he had a sound grasp of the principles of electro-physics and also reflected a large experience of his own work in the application of the method. The delightful Hunterian oration was in the historical field and proved a fine tribute to the work of Charles White, another notable Manchester surgeon. It was composed and delivered in war-time and embodied much research into local medical history.

Whatever he undertook he did with unflinching devotion to the duties and obligations involved, and his integrity and honesty were apparent. Another outstanding characteristic was his loyalty, and in that respect the Manchester School can never have had a more sincere devotee.

His principal relaxations were found in music, books, and travel. Burgess was a great family man and the death of his wife in 1941 was a lasting blow. Physically he was outstanding as a good-looking man of over six feet, of good presence, very energetic, an early riser, a quick walker, and a ready speaker, and as he got older it was borne in upon us that " youthfulness is not the prerogative of the young ". Burgess had an avidity for friendship and was always a presence for he held men together in a remarkable way, and in the surgical world will be sadly missed.

REVIEWS AND NOTICES OF BOOKS

The Treatment of Malignant Disease by Radium and X-rays. Being a Practise of Radiotherapy. By RALSTON PATERSON, *M.C.*, M.D., F.R.C.S.E., D.M.R.E., F.F.R., Christie Hospital and Holt Radium Institute, Manchester. $9\frac{3}{8} \times 6\frac{1}{2}$ in. Pp. 622 + x, with numerous illustrations. 1948. London: Edward Arnold & Co. 45s. net.

SURGEONS in general will welcome the appearance of this book because it gives them a vast amount of information on a subject in which they are of necessity interested. With the cure of most of the inflammatory conditions with the " sulpha " drugs and penicillin, the surgeon finds today that his beds are more and more occupied by cancer cases. Surgery has its limitations as far as the treatment of cancer is concerned, and the use of radium and X-rays as an adjunct to surgery is fast becoming manifest.

The Christie Hospital and Holt Radium Institute, Manchester, is known all over the world and its first and second five-year reports have stimulated all who are interested in the treatment of malignant disease.

Mr Paterson has produced a book which is lucid in style, practical, and definite to a degree. He covers those essential points, such as determination of the treatment policy, choice of technique, time and dosage, which every doctor is seeking to know. All the common sites for cancer are considered in individual chapters, and the text is illustrated by first-class photographs, radiographs, and charts.

For dealing with cancer of the cervix Dr. Margaret Tod has contributed her knowledge and experience to that of the author.

In the chapter dealing with malignant disease of the brain, the clinical preamble is written by Professor Geoffrey Jefferson and is a masterpiece by itself; the matured observation of one whose clinical and operative experience has exceeded even that of Cushing demands consideration. This so-called preamble is the best account so far published of the use of radiotherapy in the treatment of brain tumours.

Teleradium Therapy is dealt with by Professor Windeyer and J. E. Roberts and the chapter is well written and well illustrated.

The problems of protection of staff from radiation effects are fully considered and checks on the health of the staff include periodic finger inspection, regular blood-counts, and periodic dental film check on received exposure. These precautions should prove of value in all radiotherapy centres.

The medical records and statistics are given in detail and should prove a stimulus to all hospitals as to the real requirements for records and 'follow-up' of all cases.

This book portrays the Manchester method of treating malignant disease by radium and X rays and it makes interesting reading, for it is full of information on this important subject.

Recent Advances in Surgery. Third edition. By HAROLD C. EDWARDS, C.B.E., M.S., F.R.C.S., Surgeon and Lecturer in Surgery, King's College Hospital, etc. 8 × $5\frac{1}{4}$ in. Pp. 437 + x, with 131 illustrations. 1948. London: J. & A. Churchill. 24s. net.

IT is nearly twenty years since the second edition of this book was published. Much has happened since that time and surgery has undergone much progress in its many branches. The advent of the sulpha drugs and penicillin have changed the outlook of inflammatory conditions and altered in no small way the surgical treatment of these conditions.

In this third edition Harold Edwards has replaced Heneage Ogilvie as editor and it may truly be said that the new editor has upheld the tradition of the former editions. Edwards has wisely obtained the services of Brock, Northfield, and Stanford Cade, who contribute excellent sections on the thorax, the nervous system, and radiotherapy in malignant disease respectively. These sections are now such specialized subjects that the general surgeon has little time or opportunity to practice them.

The layout of this new edition has been carefully considered and the author is to be congratulated upon his selection of subjects to be included in such a volume. Much of the material cannot be obtained from other books, as it is the experience gained by the author and his surgical colleagues during the second world war.

Perhaps the best section in the book is that devoted to the alimentary canal—and this is to be expected, as Edwards has for some years spent a lot of his time and thought in the treatment of gastric lesions. The problems of carcinoma of the œsophagus, the use and abuse of vagotomy, bleeding peptic ulcer, ulcerative colitis, and carcinoma of the rectum are all considered in a practical and interesting way and the reader obtains a balanced view as to the best form of treatment.

The section dealing with the surgery of the blood-vessels is excellent and the use of heparin and dicoumarin carefully considered.

This book is a valuable contribution to surgical literature and no surgeon can afford to be without it as it gives the up-to-date considered opinion of the profession in the treatment of surgical conditions. At the end of each chapter modern references in world surgical literature enhance the value of the book and make it an important reference volume.

La Lussazione congenita dell 'anca. Nouvi criteri diagnostici e profilattico-correttivi. By MARINO Ortolani, Ferrara. 11 × 7¾ in. Pp. 129, with 118 illustrations. 1948. Bologna: F. Cappelli. Lire 1500.

IT has for long been known that the incidence of congenital dislocation of the hip in Northern Italy is very high. A great deal of the improvement in treatment since the beginning of the century derives from the Rizzoli Institute, with Putti's name at the head of a long list of Italian orthopædic surgeons who have made contributions to the advances.

It is not surprising therefore that the pædiatricians should take advantage of their early access to possible victims and seek some sign, recognizable in the first few days or weeks of life, by which the existence of an abnormal relation between femoral head and acetabulum likely to become an actual dislocation could be determined. That is precisely what Professor Ortolani claims to have done. He here describes a manipulation, applicable to the newborn babe, even if premature, which is diagnostic of a joint likely to become, or actually, misplaced. In the course of the test, a snap ("*scatto*") is felt, and sometimes "seen"; it is elicited with ease, and the procedure is quickly learnt. He says he obtains almost a hundred per cent verified correct diagnoses.

Reading the text, and looking at the numerous radiographs of joints injected either with air or an opaque substance, one feels that for conviction of the validity of the author's claim a demonstration is needed; but that, he says, is exactly what happens. Surgeons come to his clinic sceptical and leave satisfied. It is unnecessary to dilate upon the value of such a diagnostic measure, and there is evidence here that treatment begun forthwith is simple, of short duration, and successful.

Professor Ortolani has his "centre for diagnosis and prophylaxis of congenital dislocation" in a foundling hospital. His material is copious and, besides giving him great experience in treatment, has provided him with no less than thirty opportunities for anatomical study of affected joints. His text, luxuriously printed, is adorned with numerous photographs of dissections as well as radiographs of injected joints; for these alone the monograph is worth study. The author apologizes for invading the territory of the orthopædic surgeon, but recognizes that the plea is really superfluous.

From whatever source comes aid in dealing with this deformity it is welcome.

The Foot and Ankle. By PHILIP LEWIN, M.D., F.A.C.S., Associate Professor of Bone and Joint Surgery, and Acting Head of Department, Northwestern University Medical School; etc. Third edition. 9⅛ × 5⅞ in. Pp. 847, with 389 illustrations, line drawings by HAROLD LAUFMAN, M.D., F.A.C.S. 1948. London: Henry Kimpton. 55s. net.

THIS work is already well known, and its third edition is noteworthy because of the additional matter which has been produced in World War II. This consists chiefly of references to chemotherapy of wounds, to crushing and other ischæmic injuries of the foot, to rehabilitation and psychosomatic medicine. The result is a substantial enlargement of an already bulky volume which has always been interesting as an example of the modern form of specialism. Dr. Lewin is a distinguished orthopædic surgeon, but his special interest in the foot leads him into the province of the physician, in regard to neurological and circulatory disorders; of the dermatologist; and even of the psychiatrist. His ambit thus becomes so wide that in many sections the book is a compendium rather than a commentary, and the author, in his desire to include something of everything, sometimes fails to give his readers clear guidance in their own difficulties. Dr. Lewin is a master of his subject, for he has not only spent many years in the special practice of the surgery of the foot but he has gained an encyclopædic knowledge of the literature by abstracting innumerable orthopædic articles—a pursuit which has obviously influenced the style of his book, but makes it of very great value to the post-graduate student as a work of reference. Needless to say, it is beautifully and profusely illustrated.

The Surgery of the Colon and Rectum. By Sir HUGH DEVINE, M.S. (Melb.), Hon. F.R.C.S. (Eng.), F.R.A.C.S., F.A.C.S., formerly Senior Surgeon St. Vincent's Hospital Clinical School and other Stewart Lecturer in Surgery, Melbourne University; etc.; and JOHN DEVINE, M.S. (Melb.), F.R.C.S. (Eng.), F.R.A.C.S., F.A.S.C., Honorary Surgeon to Outpatients, Alfred Hospital, Melbourne. 8¾ + 5⅝ in. Pp. 362 + xii, with 277 illustrations, a number in colour. 1948. Bristol: John Wright & Sons Ltd. 52s. 6d.

THE main object of this book, as set out in the preface, is to improve the results of surgery in malignant disease of the colon and rectum, and it is clear that the authors have taken the greatest possible pains to describe and illustrate the methods which they practice. Perhaps most readers will turn first to the authors' description of the Devine defunctioning colostomy, and they will find the most perfect and clear description of this important principle, its indications, technique, and method of closure. At the present time its use in the anterior or segmental resection of the lower pelvic colon or recto-sigmoid is particularly opportune (p. 240) and references are given to C. F. Dixon's work on this subject.

The value of 'staged' methods of subtotal colectomy for polyposis and Hirschsprung's disease is shown, and the 'sliding scale' operations for ulcerative colitis with eventual ileorectal anastomosis by the use of an enterotome should be carefully studied with a view to avoiding the inconvenience of a permanent ileostomy. Many different types of conditions affecting the proximal and distal colon are described in great detail, with indications

for treatment and suitable technique for varying contingencies. Surgeons will note the description of the Devine operating frame for assisting exposure, and different types of enterotome are illustrated; the authors give very practical advice as to their use and also discuss the best times for closure of colostomies, whether early or late. A detailed description of excision of the rectum is given; for the most part the favoured method is a perineo-abdominal excision with the patient in the elevated lithotomy position and two surgeons working simultaneously, a method described by Sir Hugh Devine in 1937. A careful consideration is given both to perineal and combined methods of conservative resection of the rectum.

The earlier chapters give an erudite account of the surgical anatomy and physiology of the colon and rectum, and the final 30 pages deal briefly with hæmorrhoids, fissure, fistula, prolapse, and such like.

The book is profusely illustrated by 277 figures, many of which are in colour, and the diagrams giving details of operative procedures leave no doubt as to the methods recommended. The book covers a very wide field in extraordinary detail and is clearly based on a great personal experience. We can cordially recommend this book to the study of all abdominal surgeons and especially to those who wish to review their technique in dealing with the colon and to learn the value of the defunctioning method, which will always be associated with the name of the senior author.

The impeccable printing and the high quality and careful placing of the illustrations are such as one associates with the work of Messrs. John Wright & Sons.

Nouveau Précis de Pathologie chirurgicale. 8⅝ × 6½ in. Vol. III, Pathologie de la Tête et du Cou. By M. ARNAUD, J.-P. CALVET, L. LEGER, P. PETIT, and V. VEAU. Pp. 589, with 308 illustrations. Vol. VI, Pathologie de l'Appareil urinaire et de l'Appareil génital masculin, Pathologie de l'Appareil génital féminin. By R. COUVELAIRE, J. PATEL, and P. PETIT. Pp. 967, with 369 illustrations. Paris: Masson et Cie. Vol. III, 900 fr.; Vol. VI, 1450 fr.

THESE two volumes are under the general editorship of Professor J. Patel, but, although he is himself responsible for the section on Gynæcology, he has had the assistance of fifteen distinguished collaborators in writing the other sections.

Although the title of the symposium might lead the reader to anticipate that it is concerned only with surgical pathology, it covers in addition every aspect of the various techniques of physical examination, clinical investigation, differential diagnosis, and principles of treatment. The books are profusely illustrated with diagrams and radiographs, but the quality of the paper varies and some of the photographic reproductions on thin paper are very poor.

Volume III (pp. 577, marked 900 fr.) deals with the Head and Neck, and includes a section on congenital malformations of the face by Victor Veau. The sections on thyroid diseases and the thymus, though mentioning recent advances in thiouracil therapy and thymectomy for myasthenia gravis, discuss them in only a few lines. The section on skull injuries is on the other hand most detailed and informative, as is that on the maxilla and mandible. Poor reproduction spoils many photographs of conditions of the tongue and neck.

Volume VI (pp. 951, marked 1450 fr.) contains the sections on Urology and Diseases of Women. Professor Roger Couvelaire's section on urology is far more detailed and comprehensive than any recent publication in this country. The principles of urological investigation, both physical and instrumental, are given in the minutest detail. Nineteen pages are devoted to disorders of the bladder neck and 15 to benign prostatic hypertrophy. Millin's retropubic operation is mentioned though not in detail; 4 pages are given to the comparatively uncommon condition of spontaneous perirenal hæmorrhage. These diagrams are clear and most of the radiographs well reproduced.

The Gynæcology section of 220 pages, by Professor Patel and Dr. Petit, is mainly based on the pathology, but includes may good photographs of clinical conditions and specimens and details on hysterography, salpingography, and other accessory methods of investigation. Principles of treatment of carcinoma of the cervix are discussed in detail, but the authors appear unwilling to commit themselves and even suggest a combination of surgery and radiotherapy.

Les Embolies de l'Artère pulmonaire et des Artères des Membres. Physiologie pathologique et Traitement. By RENÉ LERICHE, Professeur au Collège de France. 9⅝ × 6½ in. Pp. 128, with 5 illustrations. 1947. Paris: Masson et Cie. 150 fr.

THIS book is the fourth in a notable series of monographs on vascular surgery which has come from the prolific pen and the fertile mind of Professor Leriche in the short space of four years, and in its preface the author gives us a clear indication that as his lecture courses survey further aspects of the surgery of the blood-vessels we may look for further additions to this library.

In dealing with pulmonary embolism he draws a sharp distinction between the large clots which form silently in the proximal veins of the lower extremities and may be deadly, and the smaller ones from manifest thrombophlebitis of the calf veins which result in comparatively less serious pulmonary infarction. For the dramatic massive embolism morphine is recommended, followed by either bilateral stellate block or the intravenous injection of novocain, either of which may combat concomitant spasm sufficiently to restore just enough of the circulation to tide the patient over the acute phase. If these measures fail Trendelenburg's operation must be performed.

For phlebothrombosis novocain block of the lumbar sympathetic trunk is first undertaken, but if œdema persists or increases incision of the main vein at the level of the groin or above is recommended for removal of the clot.

In the treatment of arterial embolism Professor Leriche hesitates to lay down definite rules because of the good results he has obtained from local arteriectomy to include the embolus, though he argues that the restoration of the circulation by embolectomy, being less mutilating, ought to give better results.

Although in every section the author refers repeatedly to the use of heparin, he points out that as they have not had any to use in his clinic his assessment of its value has been made at second hand. In reading the monograph one thus gains the impression that Leriche himself has the idea that many of his opinions might be modified if he had had some heparin at hand, and British readers should bear this consideration in mind.

Wayfarers in Medicine. Studies in the Progress and Triumphs of Medicine. By WILLIAM DOOLIN. 8½ × 5½ in. Pp. 284 + viii, with a number of plates. 1947. London: William Heinemann Medical Books Ltd. 21s. net.

Science moves, but slowly, slowly, moving on from point to point
. . . . .
Yet I doubt not through the ages one eternal purpose runs
And the thoughts of men are widened with the process of the suns.
(*Tennyson, "Locksley Hall"*)

THE history of medicine illustrates the above, probably better than that of any other science and art.

The science and art of surgery must be as old as man, in that he must have had to deal with wounds long before the time of recorded history.

The evidence of trephining in prehistoric skulls proves that the surgeon existed in the person of the witch

doctor, tribe magician, or under some other designation. It was natural that disease should have been regarded as punishment meted out by the gods, hence the office of physician and priest were one. They remained closely associated until the sixteenth and seventeenth centuries in spite of the fact that Hippocrates had freed medicine from the domination of magic by insisting that disease was due to natural rather than divine or sacred causes. It was not, however, until well after the Renaissance that the physician's role was definitely that of rendering first aid, and last aid was left to the priest. It might well be argued that by the end of the nineteenth century the separation had proceeded too far, and that the influence of mind over matter is even now not fully recognized.

It is extraordinary that the history of Medicine should have been so neglected in our medical schools and left to a few enthusiastic amateurs.

There is only one chair of the History of Medicine in the country—that of the University of Edinburgh. This compares very badly with the position in the United States of America and many European countries.

It is all the more so as clinical teachers lay such great stress on the history of a case. The reason for this is obvious. Education aims at the creation of a trained mind; a trained mind must be a discriminating one, able to distinguish between truth and falsehood, the significant and the trivial.

Diagnosis depends on physical signs and laboratory examinations; treatment on general principles whatever the disease. Little sustained effort as a rule is made by clinical teachers to instruct the student when and how those physical signs and principles were discovered and evolved.

All those interested in medicine are naturally interested in the history of their science and art. This is definitely shown by the fact that most papers always begin with a more or less accurate, even if very sketchy, review of the historical background of the condition which is to be described.

It is therefore for these reasons that such a fascinating and delightful book as this will be eagerly read and absorbed by the keen student, not merely the undergraduate but those who have been qualified many years, and especially those who have leisure to cultivate their natural tastes as they reach the twilight years of practice. The author shares this view by quoting the opinion of Fuller that " History maketh a young man to be old without either wrinkles or grey hairs " and also that of Sir William Osler, who exhorted his students to cultivate an interest in the history of medicine for three reasons: its educational worth, its association with philosophy, and its usefulness " as a pastime for the leisure moments of busy men who take an interest in the story of their profession ".

The author regards himself as a pamphleteer rather than an historian and being a surgeon his " wayfarers " tend to belong to that branch of medicine, but not wholly so. The interdependence of medicine and surgery is never overlooked, and one delightful chapter on " Descartes: The Link ", rightly coming in the middle of the book, would from its philosophical outlook have delighted Aristotle himself.

The author, though a busy surgeon, has made the leisure and has had the ability to do much besides the practice of his craft. He is the Editor of the *Irish Journal of Medical Science*, and Secretary to the Council of the Royal College of Surgeons of Ireland.

He has approached his subject through the medium of biography, agreeing with Carlyle, who said " The history of the world is but the biography of great men ", and these are not merely kings, statesmen, and politicians. The same sentiment was expressed by Emerson, who maintained that there is properly no history only biography.

Be that as it may, a perusal of this book will by way of short biographies of many of the great men of medicine, described against their proper background, widen the outlook of any reader, lay or medical. The pilgrimage he describes begins with the oldest physician and the oldest text-book, thence through medieval period along the world's trade routes. There is a long pause with Vesalius and " Leonardo The Forerunner ". After that there is naturally a diversionary visit to the art galleries of Holland to see those extraordinary pictures painted in the seventeenth century illustrating anatomical and surgical teaching, which obviously must have interested the burghers so much.

To those who have seen these pictures this chapter is an absolute joy. It is regretted that modern austerity has not allowed the publishers to make the illustrations better and larger.

The vagaries of a patient of the Renaissance in the person of Benevuto Cellini show that though medicine may have changed for the better throughout the ages, the patient has remained to all intents and purposes the same, and Montaigne is introduced as a proper contrast. The stage having been set for modern medicine by the work of Vesalius and Harvey, with " Plain Parson Hales " in the background, the pilgrimage is continued to Edinburgh, in the days of the Munros and the resurrectionists; to France with Laennec and Claude Bernard; then a trip to Canada, and thence to the Panama zone to meet William Gorgas. The pilgrim returns to this country to meet Lister, Osler, and Sir Robert Jones, with a pause, as every one should, when returning from America, for at least a short stay in Dublin. Assuredly one must stop in Dublin for so many medical movements had their genesis there. In 1710 Dr. Richard Stevens left his estate to build a hospital " for such sick and wounded, whose wounds are curable ", thus preceding John Addenbrook in Cambridge and Thomas Guy in London.

Stevens's Hospital was actually preceded by the Jervis Street one. The first hospital for incurables was founded in Dublin in 1744 and in 1745 Dr. Bartholomew Mosse founded the first hospital for women in childbirth, The Rotunda.

Medical education throughout the world owes much to Richard Carmichael, Robert Graves, and William Stokes, of Dublin. They urged the separation of the art of the apothecary from that of the physician and surgeon before the Medical Act of 1858. It was William Stokes who was President of the B.M.A. meeting in Dublin in 1867, and urged the responsibility of the State for its citizens' health and the special Diploma in State Medicine established in 1870 by Dublin University was the first in the British Isles.

The names of Graves, Stokes, Corrigan, Colles, and many others remind us of the debt medicine owes to Dublin.

The first Academy of Surgery in Europe was founded by Louis XIV in 1731— the first President was an Irishman, Georges Mareschal, whose father was an Irish Officer—one of the flock of " Wild Geese " who went abroad after the fall of Limerick and served with Sarsfield at the battle of Rocroy. This was followed by the foundation of The Royal College of Surgeons of Edinburgh (1778), Dublin (1784), London (1800).

It is little realized how much natural sciences owe to medicine. Copernicus, Galileo, Gilbert, Galvani, Boyle, Linnaeus, were all physicians. The pilgrimage of medicine in company with its wayfarers and pathfinders leads the student over the whole world.

From this book and the bibliography attached he may learn much of the route and of its travellers all seeking to probe the riddles of nature. It will be to many an ideal bedside book, but it will hardly conduce to sleep owing to its exciting story. To others after a very trying day in which failures in the science and art of their calling means perhaps anxiety and worry which prevents sleep, it will bring comfort, as it details the disappointments and frustration of others. The student

of Nature and her ways has no easy task, but the lure of the chase and the journey will be for ever there.

> And Nature the old nurse took
> The child upon her knee,
> Saying " Here is a story book
> Thy father has written for thee."
>
> "Come wander with me," she said,
> " Into regions yet untrod,
> And read what is still unread
> In the manuscripts of God."
>
> And he wandered away and away
> With Nature the dear old nurse,
> Who sang to him night and day,
> The Rhymes of the Universe.
>
> *Longfellow, " The Birthday of Agassiz " (May 28, 1857)*

Ulcers of the Stomach and Duodenum. Technique of Surgical Treatment. By EDMUNDO VASCONCELOS, M.D., F.A.C.S., Professor of Clinical Surgery, Faculty of Medicine, University of São Paulo, Brazil. 11⅜ × 8⅞ in. Pp. 125 + viii, with 154 illustrations. 1947. São Paulo: Editora Renascença S.A. Price not stated.

PROFESSOR Vasconcelos, of the University of São Paulo, has performed over 1000 partial gastrectomies for ulcer in the past twenty years and this work is a record of his personal methods of dealing with the various problems offered by different types and sites of ulcers. It is a description of operative technique only and is written for surgeons, with special reference to the XIIth Congress of the International Society of Surgery.

To the account of his own methods Professor Vasconcelos has added a short note on 100 cases of perforated ulcer, of which 85 were treated by immediate partial gastrectomy with 8 deaths, suture of the perforation being reserved for desperate cases. These operations were performed by various surgeons, mostly assistants, and represent the standard treatment of the São Paulo School.

Vasconcelos uses spinal anæsthesia with heavy percaine for all patients with a blood-pressure range between 100/70 and 140/100. Outside these limits he substitutes nerve-block of the abdominal wall, lesser omentum, and hepaticoduodenal ligament with injection of the pancreas in the neighbourhood of a penetrating ulcer.

The incision is a right paramedian one and it is kept open by a three-bladed automatic retractor. His standard operation follows the Reichel-Polya method with a horizontal section of the stomach at the level of the left gastro-epiploic artery. The duodenal stump is closed by ligature, purse-string, investing sutures, and an omental graft, and the left gastric artery is divided at the level of the section of the stomach. The Hoffmeister-Finsterer technique is reserved for ulcers high on the lesser curve. Jejunal ulcers are removed by aseptic entero-enterostomy followed by the standard resection.

The monograph is illustrated by 154 figures which show the operative manœuvres clearly in semi-diagrammatic form. It is well produced and the English translation is good.

Cancer of the Breast. By DUNCAN C. L. FITZWILLIAMS, C.M.G., M.D., Ch.M., F.R.C.S. (Edin. and Eng.), Surgeon to the Mount Vernon Hospital and the Radium Institute. 8½ × 5⅝ in. Pp. 199 + vii, with 20 plates. 1947. London: William Heinemann Medical Books Ltd. 25s. net.

THIS work expresses in a clear and practical way the long experience of Mr. Duncan Fitzwilliams on the treatment of carcinoma of the breast. Starting as a general surgeon at St. Mary's Hospital, he became attached to the more specialized Radium Institute at Mount Vernon Hospital. He is therefore in a position to speak with authority, and his conclusions, based on large numbers of case summaries and clinical training, are balanced and judicial. He describes in detail his operative treatment, and his method of treatment by means of radium alone, but regards the combination of the two as the best method for treating cancer of the breast. He therefore is an advocate for the less radical methods which were practised in Sweden between the wars, and show signs of returning to this country.

Miss Alice Ross, Radiotherapist to Mount Vernon Hospital, contributes a chapter on the X-ray therapy in operable and inoperable cases. She claims that adequate radiotherapy produces results comparable with surgery in early cases, and better results in operable but more advanced cases. Post-operatively radiotherapy confers only slight improvement in early cases, but definite improvement in later cases. She would confine surgery to the outer half of the breast.

The average general surgeon, who is less fortunate in commanding these technical and scientific advantages, should nevertheless glean much information from this book for improving his treatment, because radical mastectomy is still the yardstick by which the results are measured in this unsettled and highly debatable subject.

Sir Frederick Banting. By LLOYD STEVENSON, M.D. 9⅛ × 6⅛ in. Pp. 446 + xvi. Illustrated. 1947. London: William Heinemann Medical Books Ltd. 25s. net.

" The art of biography appears to have fallen on evil times in England. Those two fat volumes with which it is our custom to commemorate the dead . . . who does not know them with their ill-digested masses of material, their slipshod style, their tedious panegyric, their lamentable lack of selection, of detachment of design.

" They are as familiar as the *cortège* of the undertaker and wear the same air of slow funereal barbarism."—(Lytton Strachey.)

THIS book cannot be stigmatized thus: Lytton Strachey would have enjoyed this biography. It is more than a documentary compilation of facts. It is intensely human and, like a portrait, depicts the whole man in all his phases rather than a series of photographic impressions which portray him at various moments of time.

There is little criticism or judgement on the part of the author; he just exposes the whole man in his development from student to maturity.

It is summed up in the words of Dr. G. E. Hall, the Dean of Medicine at the University of Western Ontario, who writes the foreword: " Banting was above all a great man, respected for his abilities, loved for his weaknesses, admired for his humility, and venerated for his achievements. Not a great scientist, as scientifically trained people appreciate the word, he was primarily a stimulator of science and scientific thought—a symbol of medical research ". This may not be an acceptable criterion to *Researchers* with a capital R, but many advances in medicine have been achieved by what appears to have been fortuitous chance, which is more often the flash of genius which recognizes the obvious. Is it possible to be trained in research or is it only possible to initiate the novice in the pathways?

A biographer need not tell all he knows. He must choose what is essential and convey to the reader an idea of the whole man—what he really was, what he looked like, the tone of his voice, and the style of his conversation. The author does this and so attains a truthful transmission of personality.

Frederick Banting, the grandson of an Englishman, was born on November 14, 1891, at Alliston, Ontario. He chose medicine for a career, but from the examination point of view he was not a brilliant student. " His method was not a quick and shallow assimilation exhibited in fluent recitals from books; but a slow milling over in his mind of everything which was offered him for study;

some of it he retained, some of it he forgot, most of it at any rate he thought about". Would that there were more such students! How easy would be the work of examiners.

He became qualified and joined up as soon as war was declared in 1914.

On September 27, 1918, he was wounded, but refused against orders to go back to the C.C.S. and though suffering from shock and hæmorrhage he remained on duty with his battalion throughout the battle until the next day. He was awarded the Military Cross for his devotion to duty. He also refused later to allow the surgeons in England to amputate his right arm on account of sepsis and made a complete recovery. During his stay in England he assisted Dr. Bruce Robertson in his pioneer work on blood transfusion. He also became keenly interested in orthopædic surgery, and it was this which prompted him on his return to Toronto to enter the Children's Hospital as a resident in the service of the late Mr. G. L. Starr. It was while experiencing the boredom of waiting for practice that Banting stumbled, so to speak, against the idea which led to the discovery of insulin and the control of diabetes. In 1869 Paul Langerhans, while still a medical student, discovered the structures which were later named after him by Laguesse. A year after Langerhans died (1889) Von Mehring and Minkowski proved that removal of the pancreas in some animals invariably produced diabetes and death.

On October 20, 1920, Banting read an article in *Surgery, Gynecology and Obstetrics* on "The Relation of the Islets of Langerhans to Diabetes with Special References to Cases of Pancreatic Lithiasis" by Moses Barron, of Minneapolis.

His reasoning can be more or less followed by the following words written in a note book about 2 a.m. on October 21, 1920. "Tie off pancreas ducts of dogs. Wait six or eight weeks. Remove and extract."

There is the idea, which germinated and fructified, and ultimately was the means of controlling diabetes. On January 11, 1922, the first patients were treated with the extract at the Toronto General Hospital.

There were no toxic effects such as occurred in 1908 when Zuelzer and his associates reported their results in the treatment of six cases of diabetes with the pressed juice of the pancreas extracted with alcohol and evaporated to dryness.

In August, 1922, the first amputation for diabetic gangrene was done after treatment with insulin and no coma developed. Banting had conceived the idea, though it had also occurred earlier to others, but as Oliver Wendell Holmes said "many ideas grow better when transplanted into another mind than in the one where they spring up". But even so, Banting always insisted that the discovery of insulin was the result of team work and not due to himself alone. Nor did he ever claim that insulin cured diabetes. It merely controlled it, or in other words it is a working cure, just as in 1926 George R. Minot, who had himself been treated with insulin for diabetes since 1922, and W. P. Murphy discovered a working cure for pernicious anæmia.

The whole book is fascinating reading and stirs the imagination. It enables the whole man to be appreciated. It has been said that a biographer never admits his Achilles has a vulnerable heel, yet without that heel Achilles would not have been a hero. This biography freely admits the heel, and uses it to demonstrate the man. As he got older, Banting's general interest in and usefulness for his fellowmen widened. He always endeavoured to fulfil his own requirements for a research worker "with self detachment, self abandonment and self abnegation, he must pursue his own ideals and ideas. Above all he must work with integrity, intensity, breadth, patience, thoughtfulness and faithfulness".

Honours were heaped upon him: The Nobel prize was shared between him and Professor Macleod in 1923. In 1934 he was honoured with the K.B.E., and in 1934 he was made a Fellow of the Royal Society. On September 16, 1930, The Banting Institute for Medical Research in the University of Toronto was opened with Banting as the first Director. He was an ideal director, more interested in the originality of those who asked his help than in their encyclopædic factual knowledge. When the war came he was deep in research, especially with regard to cancer and silicosis.

All was dropped when the war came; he immediately applied for war employment and devoted himself to the elucidation of the enigmas of aviation, physiology, and medicine. The end came on February 20, 1941, when the plane in which he was to cross to England had to return to base on account of engine trouble and was destroyed in making a crash landing at Musgrave Harbour in Newfoundland. The description of his departure from Gander Bay in Newfoundland to the finding of his dead body is absolutely thrilling and realistic. So he died, as he had lived—in the midst of working and thinking out ideas to help others.

The book is one which should be included in all school libraries alongside those recording the life of Scott, Shackleton, and innumerable others which record how a man should live and die. No book could be better as a prize to the so-called best student of his year; it would make him not only a better student, but a better man. His memory will remain, as in the words of Lord Moynihan at the opening of the Banting Institute:

"Memorials are of many kinds, some are graven in stone, some are cast in bronze, some are written in letters of gold on the roll of honour which enshrine the names of the saviours of their country. There is yet another memorial, born in the hearts of those from whom a heavy load of suffering and of sorrow has been lifted, and dedicated by them as an act of homage to a great deliverer."

La Chirurgie biliaire sans Controle manométrique et radiologique per-opératoire. By PIERRE MALLET-GUY, RENE JEANJEAN, and PIERRE MARION. 12¾ × 9¾ in. Pp. 140, with 170 illustrations. 1947. Paris: Masson et Cie. 1200 fr.

THE authors give a detailed account of their technique for studying the biliary tract during the course of an abdominal operation. For this purpose they insert a cannula into the gall-bladder or, if this has been removed at a previous operation, into the common bile-duct. A manometric curve of the pressure is recorded on a rotating drum for 25 seconds, after which the gall-bladder or ducts are filled with an opaque medium and radiographed. When the injection is into the gall-bladder, they use "ténébril"; when into the duct, iodized oil. They use a simple operating table with a radio-translucent top and without a central pillar, so that the X-ray tube can be placed underneath the table. When the radiographs are taken, a sterile towel is placed over the field of operation, and the film rapidly drawn across until it is directly over the gall-bladder; a simple centring device ensures that it is correctly orientated to the X-ray beam. One radiograph is taken first and quickly developed (in three minutes), after which a second radiograph is taken.

After defining the normal manometric curves and normal X-ray appearances produced by this examination, the authors proceed to describe a condition of hypertonia of the sphincter of Oddi, which they claim to be clearly demonstrable both on the pressure curve and on the radiographs, and which they treat by a transduodenal operation on the sphincter. They are also able to demonstrate a condition of hypotonia of the gall-bladder and ducts; they treat this by resection of the splanchnic nerves. This approach to biliary dyspepsia is certainly provocative and will need confirming by further work

in this country. The clinical evidence given in this book is not sufficiently detailed for the validity of the theory to be assessed.

The procedure is of undoubted value, however, in cases of calculi and inflammation, when it is important to ensure that the ducts are normal as a prelude to the removal of the gall-bladder. If the conditions of hypertonia or hypotonia are found to be present, the authors deal with these at the same time as the removal of the gall-bladder. In cases in which the gall-bladder has been removed at a previous operation, such cholangiography during the course of a second operation is of undoubted value. It is also useful for studying the condition of the bile-ducts in chronic pancreatitis.

The book is profusely illustrated with reproductions of excellent radiographs, each accompanied by a diagram clarifying the appearances shown, and, in most cases, by a reproduction of the relevant pressure curve. A short description of the clinical history and of the operative procedures adopted for treatment is given with each set of illustrations. The work should certainly be studied by all surgeons interested in the surgery of the biliary tract.

Pictorial Handbook of Fracture Treatment. By EDWARD L. COMPERE, M.D., F.A.C.S., and SAM W. BANKS, M.D., F.A.C.S., Northwestern University Medical School. Second edition, revised with the assistance of CLINTON L. COMPERE, M.D., F.A.C.S. Illustrations by HAROLD LAUFMAN, M.D., F.A.C.S. 8 × 5½ in. Pp. 390, with 191 illustrations. 1947. Chicago: The Year Book Publishers Inc. (London: H. K. Lewis & Co.). 30s. net.

THIS little book—now in its second edition—has a great deal to commend it. Undoubtedly the outstanding feature is the series of line drawings by Dr. Laufman—artist and surgeon. These serve to illustrate almost every paragraph of the text and are remarkably clear. Combined with many excellent radiographs and diagrams, they fully justify the label "pictorial."

The scope of the book is comprehensive—certainly one would imagine beyond the sphere of the general practitioner for whom the book was primarily written. But for the senior student or resident medical officer it would offer valuable pabulum both from the examination and practical viewpoints.

The authors—orthopædic surgeons of long experience—wisely adhere to their originally expressed intention of describing for each lesion one well-tried and proven method of treatment rather than offering a series of theoretical possibilities. The result is a book which, though essentially a summary, is readable and pictorially tempting. It is interesting to note in how few cases the treatment advocated differs in principle from that accepted in this country, at any rate for the commoner fractures and injuries. The details are obviously personal, often intriguing, and not dangerous.

In these days of economic stress and strain it is refreshing to find the emphasis that is laid on rehabilitation—not only in the after-treatment of recent fractures but in dealing with the loss of function which is the aftermath of the old maltreated or malunited fracture.

In all a pleasant book—practical, pithy, and predominantly pictorial.

Neurological Complications after Spinal Anæsthesia. And Results from 2493 Follow-up Cases. By GUNNAR THORSÉN. Being Supp. 121, vol. xcv, *Acta chirurgica Scandinavica*. Translated by ULLA SCHÖTT. 9⅛ × 5¾ in. Pp. 272, with 11 figures and 5 plates. 1947. Stockholm: P. A. Norstedt & Söner. 15 kr.

THIS is a comprehensive survey of the literature dealing with the complications of spinal anæsthesia and the results from 2493 follow-up cases.

Any surgeon or anæsthetist reading this monograph will probably never suggest the use of a spinal anæsthetic again, for the complications appear to be legion, ranging from persistent dura fistula to manifest cerebral lesions with epileptic fits, pareses, hemiplegias, and changes in consciousness, personality, and tonus. Probably the commonest complication due to spinal anæsthesia is headache, which at times may be permanent.

This book gives the reader the very definite impression that a number of nervous sequelæ are completely overlooked after spinal anæsthesia.

Confrontations radio-anatomo-cliniques. Edited by De M. CHINAY, R. A. GUTMANN, and J. SÉNÈQUE. Fasc. II. 12¾ × 9¾ in. Pp. 70, with 140 illustrations. 1947. Paris: G. Doin et Cie, Masson et Cie. 465 fr.

THIS book contains a number of case histories by different authors, in which emphasis is placed on the radiological findings. The cases include various bone conditions and numerous lesions of the stomach, colon, and kidneys, some of them being rare and some fairly common in this country. The text is illustrated by well-reproduced radiographs, in which the relevant X-ray appearances are for the most part clearly shown.

There is no systematic selection of the cases, and it is rather difficult to see the purpose of the book, though parts of it, such as the sections on tuberculosis and lymphosarcoma of the stomach, and some of the rarer bone tumours, have value as a museum of rare conditions.

Terapia endoarteriosa. By E. MALAN and G. ENRIA. With a Foreword by Professor A. M. DOGLIOTTI. 9⅝ × 6¾ in. Pp. 184 + vii, with 44 illustrations, a number as plates. 1947. Turin: Minerva Medica. L. 900.

THE research reported in this monograph began before penicillin became available, and suffers in value in consequence. It was an attempt to deal particularly with localized or spreading inflammation by bringing antiseptics into more intimate and perhaps more prolonged contact with the affected tissues than had proved possible by other routes. The authors are now reduced to supporting the proposition that even in the case of so efficient a remedy as penicillin there may be advantages.

When one looks dispassionately at the records of their attempts with flavine, gentian violet, and mercurochrome, which give severe reactions both functional and organic in the arterial, arteriolar, and even capillary systems, at their thromboses and obliterations, the apologies for recoverable incidents and severe pain ring rather dismally in one's ear. It would call for very striking demonstration of superiority to other routes and other methods to justify the hazards, even though they may be rare with suitable materials in skilled hands.

As a method for administration of insulin, or for combating œdema, there is little to be said even by those who believe in or employ it. All this is not to say that the technique of arterial puncture should not be learnt, or that there may not be exceptional occasions for this form of therapy. The book at least documents the risks.

L'Infiltration stellaire. Technique, Indications, Résultats. By G. ARNULF, Nancy. With a Preface by Professor R. LERICHE. 8¾ × 6½ in. Pp. 234, with 25 illustrations. 1947. Paris: Masson et Cie. 400 fr.

THIS book belongs to a series, almost a library, of volumes dealing with the disorders of the vasomotor system, which have been written by Professor Leriche and his pupils and associates. Professor Arnulf acknowledges his indebtedness to the inspiration and example of his Master, who has written the preface to this book.

Clinicians and also physiologists who are interested in the autonomic nervous system will find much valuable information in this volume, which gives a fully documented account of the anatomy of the stellate ganglion and the technique of blocking it by injection.

The greater part of the work, however, consists of detailed clinical records of the very numerous conditions for which stellate injection has been performed, and in every case the physiological basis for the procedure and the results obtained are carefully examined.

The scope of the work is astonishing, for the author has aimed at completeness rather than at the selection of the conditions for which stellate block is of recognized value. But all the information is here—the clinician can decide for himself whether the procedure seems to him to be justifiable or not.

BOOK NOTICES

[*The Editorial Committee acknowledge with thanks the receipt of the following volumes. A selection will be made from these for review, precedence being given to new books and to those having the greatest interest to our readers.*]

Emergency Surgery. By HAMILTON BAILEY, F.R.C.S., F.A.C.S., F.I.C.S., F.R.S.E., Surgeon and Surgeon-in-charge of the Genito-urinary Department, Royal Northern Hospital, London; etc. Sixth edition. 10 × 6½ in. To be issued in five parts. Part I, pp. 1–180 + viii, with 289 illustrations, a large number in colour. 1948. Bristol: John Wright & Sons Ltd. 21s. per part, binding case supplied with Part V.

Surgical Treatment of the Abdomen. Edited by FREDERIC W. BANCROFT, A. B., M.D., F.A.C.S. Associate Editor: PRESTON A. WADE, A.B., M.D., F.A.C.S., Associate Professor of Clinical Surgery, Cornell University Medical College. 10 × 7 in. Pp. 1026 + xviii, with 457 illustrations and 3 coloured plates. 1948. Philadelphia and London: J. B. Lippincott Co. £5 10s. net.

Loose-leaf Diagnostic Roentgenology. Edited by ROSS GOLDEN, M.D. Third issue of renewal pages. 1948. New York and Edinburgh: Thomas Nelson & Sons.

The Practice of Local Anæthesia. By GEORGE BANKOFF, M.D., D.Ch., F.R.A.P.S., F.R.C.S.E., Surgeon, Italian Hospital; etc. Third edition. 8½ × 5½ in. Pp. 290, with 199 illustrations. 1948. London: Staples Press, 30s. net.

Radon: Its Technique and Use. By W. A. JENNINGS, B.Sc., A.Inst.P., Physicist to the Royal Northern Hospital and Prince of Wales's General Hospital Joint Radiotherapy Centre; and S. RUSS, C.B.E., D.Sc., F.Inst.P., Formerly Physicist to the Middlesex Hospital, London. 8¾ × 5⅞ in. Pp. 222 + x, with 14 plates and 49 illustrations in the text. 1948. London: John Murray, for the Middlesex Hospital Press. 18s. net.

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